

Running head: SOCIAL SUPPORT AND STIGMA IN MS AND MND

Submitted in partial fulfilment of the Lancaster University Doctorate in Clinical Psychology



Doctoral Thesis

**The influence of a lack of social support and
perceived stigma for individuals with multiple
sclerosis and motor neurone disease**

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SOCIAL SUPPORT AND STIGMA IN MS AND MND

Word Count Statement

Thesis Section	Main Text	Appendices (including title pages, abstracts, tables, figures & references)	Total
Thesis Abstract	300	-	300
Literature Review	7,750	12,774	20,524
Empirical Paper	7,584	7,583	15,167
Critical Appraisal	3,973	1,200	5,173
Ethics Section & Appendices	5,693	4,478	10,171
Total	25,300	26,035	51,335

Thesis Abstract

Section 1 describes a systematic literature review examining psychological correlates of perceived social support in multiple sclerosis. Five subject databases (CINAHL; EMBASE; PsycINFO; PubMed; Web of Science) were searched using a single search string (correlates OR factors AND “social support” and “multiple sclerosis”). Of the 21 articles reviewed, 20 reported statistically significant relationships. Most studies ($n = 11$) correlated perceived social support with depression or mental health aspects of quality of life and all found the higher the social support, the lower depression and higher positive mental health. Studies using regression identified that greater social support was a significant predictor of lower anxiety, anger, depression, loneliness and better mental aspects of health-related quality of life, postpartum emotional distress and self-esteem. The results provide evidence for significant relationships between social support and various psychological variables.

Section 2 describes a research study which aimed to identify whether a lack of social support and increased levels of perceived stigma predicted psychological distress for individuals with motor neurone disease (MND). Correlational and hierarchical regression analyses were conducted. Significant correlations were identified between social support, felt and enacted stigma and measures of psychological distress. Regression analyses revealed that enacted stigma was not an independent predictor in any of the models and social support did not remain a significant independent predictor for stress when stigma entered the model. Moreover, felt stigma was a more powerful significant independent predictor in all the models. It may be important to consider social support and stigma when aiming to improve psychological distress for individuals with MND.

Section 3 describes a critical and reflective appraisal of aspects of the whole thesis. This includes an overview of the main findings, discusses recruitment issues, the

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conceptual framework of disability employed to guide the terminology usage, and makes recommendations for future research.

Declaration

This thesis records research undertaken for the Doctorate in Clinical Psychology programme at the Division of Health Research at Lancaster University from October 2017 to September 2019. The work presented here is the author's own except where due reference is made. The work has not been submitted for the award of any higher degree elsewhere.

Natalie Leigh

13/09/2019

Acknowledgements

Firstly, I would like to thank all the individuals who took part in this research, they selflessly gave their time to participate, without which this research would not have been possible. I would also like to thank the various organisations and individuals for sharing the study details and supporting the advertisement and recruitment of this study, their assistance was invaluable.

I would also like to thank my Research Supervisor, Fiona Eccles, and my Field Supervisor, Jane Simpson for being incredible supervisors throughout this process. Their unwavering support and guidance are greatly appreciated and helped me through the most difficult times.

Many thanks to the member of Lancaster University Public Involvement Network who provided their time to review the study documents and provided their input with regards to the design of the study.

Finally, I would like to thank my friends, family and fellow trainees who have supported me in so many ways throughout this process. Particularly, my sister-in-law for her insights into the experiences of individuals with MND and their family members, along with her support in reviewing the study documents. I would also like to thank my sister and dad, for their encouragement and for always believing in me, my mum and husband for their constant faith and for picking up all the childcare and household tasks when required. Most importantly my son, for giving me the motivation to keep going.

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Section 1: Literature Review

Psychological correlates of perceived social support in multiple sclerosis: A systematic review

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Word count (exc. title page, abstract, appendices, figures and tables): 7,750

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Abstract

Purpose: Social support has been identified as a significant correlate and predictor of variables relating to psychological distress and well-being in neurological conditions. This review aimed to discover if perceived social support was a significant correlate and predictor of psychological variables for individuals with multiple sclerosis (MS). The definition of psychological variables adopted for this review related to psychological distress, well-being and concepts such as depression, self-esteem, mental aspects of health-related quality of life and other mental health outcomes. **Methods:** A systematic literature review was conducted using five electronic databases and a single search string. Articles were systematically screened and those which met the inclusion criteria were included in the review ($n = 21$).

Results: Of the 21 articles reviewed, 20 reported statistically significant relationships. The correlations with the largest effect size were found between social support and depression, hope and mental composite of quality of life, whereby higher levels of social support correlated with lower depression and increased hope and better mental-health related quality of life. The variable with the largest number of studies and significant results was depression, followed by mental aspects of health-related quality of life. Regression results identified that greater social support was also a significant predictor of lower anxiety, anger, depression, loneliness, and better mental aspects of health-related quality of life, postpartum emotional distress and self-esteem. **Conclusions:** The results provide evidence for significant relationships between social support and various psychological variables. However, more robust research is required to improve the quality of these findings.

Keywords: Social support, depression, quality of life, correlates, multiple sclerosis

Introduction

Social support is a complex concept, with varying definitions (Berkman, Glass, Brissette & Seeman, 2000). For example, it has been described in terms of a social network structure, often measured quantitatively (Hirsch, 1980; Hutchinson, 1999), which is comprised of a set of significantly present others through which social support is gained. A sociological perspective provided by Durkheim (1951), proposed that membership of social groups can create a sense of belonging and provide meaningfulness to an individual's life, and that lack of socialisation can lead to hopelessness and despair. This suggests that individuals' level of integration within their social network can directly affect their psychological well-being. However, it has also been argued that being embedded in social networks can also affect both positive and negative outcomes. For example, Burg and Seeman (1994) reported that a larger social network size does not always provide positive health outcomes for individuals with physical health conditions, as extensive networks can produce costs as well as benefits. Furthermore, Chak (1996) criticised using social network as a conceptualisation for social support because this assumes that all social interactions are helpful. Alternatively, satisfaction with social support, rather than the size of the social network may be important and, was emphasised theoretically in Lazarus and Folkman's transactional stress model (1984), which proposed that the individual's evaluation of the support as being adequate was vital to avoid distress.

A further conceptualisation is that of perceived social support. This concept was first theorised by Sarason and colleagues (Sarason, Sarason & Shearin, 1986; Sarason, Pierce & Sarason 1990; Sarason et al., 1991) and is considered one of the most well researched concepts of social support (Chronister, Johnson & Berven, 2006). This conceptualisation suggests that how social support is perceived by the individual is most important. While similar to satisfaction with social support, it differs in that perceived social support does not

focus entirely on the levels of satisfaction with that support, rather, it refers to the individual's perception of support (i.e. perceived levels/ amount of support received, perceived satisfaction with support received, perceived sources of support etc.) Perceived social support is useful in that it is specific and has been widely used in both research in this field and as a construct within most definitions and measures of social support (Chronister et al. 2006; Chiu, Motl & Ditchman, 2016). As such, this understanding of social support will be used within this quantitative systematic literature review.

The way in which social support is related to health has been theorised by Heaney and Israel (2008) who hypothesise that social support is the starting point of a causal flow to health outcomes. They state that, in actuality, some of these relationships could be reciprocal as health status can influence the extent to which an individual can maintain and recruit social support. In this model, social support can influence health behaviours through a direct pathway by impacting physical, mental and social health or through indirect pathways of individual coping resources or organisational and community resources. Moreover, this model will be used as a theoretical framework to guide the results of this review and to illustrate this conceptualisation of the relationship between social support and psychological factors.

Perceived social support has been useful in research investigating psychological distress and well-being across a wide range of physical health conditions such as women at risk of hereditary breast cancer (den Heijer et al., 2010); Parkinson's disease (Saeedian et al., 2014; Garlovsky, Overton & Simpson, 2016); individuals accessing cardiac care (León-Pérez, Wallston, Goggins, Poppendeck & Kripalani, 2016; Sirri, Magelli & Grandi, 2011); fibromyalgia (van Koulil, van Lankveld, Kraaimaat, van Riel & Evers, 2010), rheumatoid arthritis (Evers, Kraaimaat, Geenen & Bijlsma, 1997); motor neurone disease (Matuz, Birbaumer, Hautzinger & Kübler, 2010; Matuz, Birbaumer, Hautzinger & Kübler, 2015) and

epilepsy (Gandy, Sharpe & Perry, 2011). One condition where social support is particularly important is multiple sclerosis (MS), a progressive autoimmune condition. Research has identified that as an individual's level of impairment increases (some of which can be due to the limitations and barriers which society impose; Simpson & Thomas, 2014), more support may be required. As people with MS tend to be of working age, support needs may change over time.

The effects of MS vary between individuals and can be difficult to predict, as the effects and severity can vary over time. MS can affect multiple sites across the central nervous system including the cerebral hemispheres, optic nerve, spinal cord and brainstem. There are five subtypes of MS, which include relapsing remitting, secondary progressive, primary progressive, progressive relapsing and benign; with relapsing remitting being the most common (Malik, Donnelly & Barnett, 2014).

Psychological difficulties and their management are important aspects of holistic care for individuals with a diagnosis of MS and extensive research has been conducted in this field. Many studies have been conducted in relation to depression (McIvor, Riklan & Reznikoff, 1984; Mohr, Classen & Barrera, 2004; Patten, Metz & Reimer, 2000) and it has been reported that this is the most common psychological difficulty (Minden & Schiffer, 1993). Other psychological difficulties have also been explored, such as agitation, irritability, apathy, euphoria, disinhibition, hallucinations and delusions (Diaz-Olavarrieta, Cummings, Velazquez & Garcia de al Cadena, 1999); anxiety (Diaz-Olavarrieta et al., 1999; Korostil & Feinstein, 2007); mood, fatigue, self-efficacy (Motl, McAuley, Snook & Gilotoni, 2009); self-esteem (Foote, Piazza, Holcombe, Paul & Daffin, 1990; O'Brien, 1993); and quality of life (Motl et al., 2009). As the above studies outline, psychological difficulties can be common in individuals with a diagnosis of MS and while a number of factors contribute to

this, the contribution of social support is an important determinant to review, especially given its potential to be changed.

A quantitative approach was taken for this review as the vast majority of research conducted in this area utilises quantitative methods, which mainly assess statistical relationships between predictive and outcome measures. Although qualitative research has been conducted on social support, this mainly focuses on the relationship between social support and physical aspects of the condition such as exercise and fatigue (Carroll, Chalder, Hemingway, Heyman & Moss-Morris, 2016; Christensen, Brincks, Schnieber & Sørensen, 2015; Christensen, Brincks, Schnieber & Sørensen, 2016; Chiu, Bezyak, Griffith & Motl, 2016; Dlugonski, Joyce & Motl, 2012).

Previous reviews have been conducted which have focused on depression and MS (Arnett, Barwick & Beeney, 2008) and psychosocial correlates of adjustment in MS (Dennison, Ross-Morris & Chalder, 2009). There is some overlap between these reviews and the current review, as perceived social support was included as a psychosocial factor within the Dennison et al. (2009) review and as a correlate of depression in the Arnett et al. (2007) review. However, these reviews were conducted at least 10 years ago, and social support was not the focus of these reviews, rather one of several components. Furthermore, the way in which the Dennison et al. (2009) review was theorised differs from the current review, as they were seeking predictors of adjustment outcomes, whereas the current review was seeking research investigating social support and its relationship with other psychological variables. Therefore, the current review aims expand on these, by identifying more recent research and by assessing the specific concept of perceived social support.

This systematic literature review therefore aims to provide an overview on the research to date that has investigated the relationship between perceived social support and

psychological outcomes relating to well-being and distress for individuals with a diagnosis of MS. Adopting a broad definition of psychological correlates has advantages and disadvantages; one advantage of such a definition is the alignment with previous literature on adjustment. Moreover, this definition provides an original, comprehensive review of the impact of perceived social support on emotional and behavioural psychological factors for people with MS. However, by adopting this definition, other variables which relate to cognitive or neuropsychological functioning, or which could be conceptualised from either a physical or psychological perspective, such as pain and fatigue, were not included. Furthermore, by including a range of disparate factors which have been found to be correlated with social support necessitates the need to consider the results within an overall conceptual framework.

Despite a wealth of literature being available on the influence of social support on such outcomes, no systematic review has been completed in this area previously. Consequently, this quantitative systematic literature review aims to enhance the evidence base by assessing the robustness of relationships between psychological variables and perceived social support, so that appropriate interventions can be identified, drawing upon theories of social support. This includes studies which used social support as a predictor (independent variable) or an outcome (dependent variable) as both of these theoretical perspectives capture relationships of interest.

Method

Search Strategy

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA; Moher, Liberati, Tetzlaff & Altman, 2009) checklist was used to guide the structure of this review (see Appendix 1-A). Consultation with a subject-specific librarian took place prior to

searches being undertaken to maximise the efficiency of the searches. Five individual subject databases were searched electronically using the single free text search string: correlates OR factors AND “social support” AND “multiple sclerosis”. Each subject database was searched individually using the above single search string. The databases used for this search were CINAHL (Cumulative Index to Nursing & Allied Health), EMBASE (Excerpta Medica), PsycINFO, PubMed and Web of Science. These databases were selected as they were the most relevant ones for health, neurology, psychology and medicine at the time the searches were conducted.

The search string used was selected as it encompassed all the aspects of the search in a succinct way, yielding the maximum relevant results. No date exclusion criteria were set for the searches and these were conducted on 5th February 2019, so articles published up to this date were included. The inclusion criteria were that the article had to investigate psychological correlates of perceived social support with individuals with a diagnosis of MS (including self-report of diagnosis). Also, both the measures of social support and the psychological variables had to be shown to be statistically valid and reliable either by the study using the measure or by previous research. Psychological correlates were defined as: “potentially modifiable factors relating to the individual’s attitudes, thoughts, feelings, and behaviours that would be relevant and possible to address in a psychological intervention” (Dennison et al., 2009, p.142). Articles were excluded if they were: not published in English, qualitative articles, review articles, conference papers, commentary articles, presentation abstracts or articles which investigated family members/ caregivers of an individual with a diagnosis of MS. Articles were also excluded if they used a different construct of social support. Thus, articles were excluded which did not investigate perceived social support; did not include an emotional support construct; looked at marital/ partner support rather than a broader concept of social support; the construct of social support related specifically to

function or work. Finally, if a quality of life measure was used then this had to provide separate scores for physical and mental components of quality of life, as only the mental components were included in the review. These criteria were used to ensure that only studies investigating psychological correlates, using a concept of perceived social support, with individuals with a diagnosis of MS were included in the review. To be as inclusive as possible and to gain a broader view of the topic, there was no restrictions on age of participants or location of the research.

The initial search was conducted using the search string and databases outlined above, the duplicates were removed, and title screening was conducted to exclude any irrelevant articles. Screening was continued using the article abstracts and any articles that did not meet the inclusion criteria or met the exclusion criteria were removed. The same process was followed for remaining articles using the full-text. Following the different stages of screening, 19 articles remained from the initial searches, and a further two articles were identified through snowballing from these articles, resulting in a total of 21 articles included in the systematic review (see Figure 1 for a flow diagram outlining this process).

The type of studies included in this review reported correlational and regression analyses that measured relationships between perceived social support and psychological variables (as defined above) in individuals with a diagnosis of MS. Studies were included if they used social support as a predictor (independent variable) or an outcome (dependent variable) as both theoretical perspectives capture relationships of interest. For the purpose of this review, pain and fatigue were not considered psychological correlates, as these were reported in terms of physical health and physical activity within the studies (Motl, McAuley, Snook & Gilotoni, 2009; Osborne et al., 2006; Osborne, Jensen, Ehde, Hanley & Kraft, 2007) and were therefore categorised as a physical correlate and excluded from the review.

The method by which these variables were collected included self-report and clinician administered measures.

Insert Figure 1 here please

Quality Assessment

Quality assessments were conducted to assess the quality of each article, and to provide a framework for the results to be viewed, the quality assessment was not used to exclude articles. An amended version of the NICE (National Institute for Health and Care Excellence) quality appraisal checklist – quantitative studies reporting correlations and associations (2012; see Appendix 1-B) was used for this purpose. This tool was specifically developed for assessing correlational studies and is based on the appraisal step of the ‘Graphical appraisal tool for epidemiological studies (GATE)’ developed by Jackson et al. (2006). This tool allows the reviewer to rate the study’s internal and external validity by viewing the key aspects of the study design (participants’ characteristics, definition of independent variables, outcomes assessed and methods of analyses). The tool includes five sections, with section one relating to external validity and sections two to four relating to internal validity with the fifth section providing a summary of scoring for the external and internal validity. There are 17 questions spread over the first four sections and each is rated on a scale, with five possible responses (see Appendix 1-C for scoring descriptions). For the purposes of this review, only 16 of the questions were used, with question 2.5 being omitted, as this question related to the applicability of the setting to a UK sample, which is not relevant for this review. For this review, one main reviewer assessed each study, and 25% of

the studies ($n = 6$) were reviewed again by a second reviewer (psychology graduate, undertaking a doctoral clinical psychology programme). The reviewers then cross-checked the ratings and any discrepancies were discussed, and a rating mutually agreed, to enhance their reliability. The results of the quality assessment were used to enhance the findings of the review, by providing a framework to view the quality of the individual study's findings. The quality assessment results were taken into consideration when drawing conclusions about the strength of the findings relating to specific variables, to either consolidate or dispute the conclusions drawn. See Table 1 for the outcome of the quality assessment ratings.

Insert Table 1 here please

Results

Of the 744 identified articles, duplicates were removed leaving 435 articles; these were screened by title and irrelevant articles were removed ($n = 233$). This left 202 articles which were then screened by abstract to see if they met the inclusion or exclusion criteria. Records were excluded if they did not meet the inclusion criteria ($n = 149$). The remaining 53 articles were assessed for eligibility using the full-text and articles that did not meet the inclusion criteria or met the exclusion criteria were excluded ($n = 32$; see Appendix 1-D for list of articles and reasons for exclusion). This resulted in a final total of 21 studies which were included in the overall review (see Figure 1).

Study Characteristics

The main study characteristics of the 21 reviewed studies can be found in Table 2. The date range for the studies was from 1984 until 2018, with the majority being conducted

in the past 11 years ($n = 11$). Thirteen studies (62%) were conducted in the USA, with the remainder being completed in Australia (McCabe, McKern & McDonald, 2004), Egypt (Effat, Azzam, Shalash, Elkatan & Elrassas, 2016), France (Gay, Vrignaud, Garitte & Meunier, 2010), Israel (Schwartz & Frohner, 2005), Lebanon (Farran, Ammar & Darwish, 2016), Poland (Jaracz et al., 2010), Portugal (Costa, Sá & Calheiros, 2012) and Saudi Arabia (Hyarat, Al-Gamal & Rama, 2018).

Despite the intended inclusion of studies which used social support as either a predictor (independent variable) or an outcome (dependent variable), all the studies identified in this review theorised social support as a predictor (independent variable) and the psychological variables as outcomes. Most studies ($n = 19$) were cross-sectional single time point studies, apart from Gulick and Kim (2004) and Koelmel, Hughes, Alschuler & Ehde (2017) which both included a longitudinal element to their research design.

The type of participants also varied across the studies, with all articles including individuals with a diagnosis of MS, which was assessed through self-report, physician confirmed, laboratory confirmed or confirmed through medical records. Some studies were aimed at specific MS populations, such as veterans with a diagnosis of MS (Bambara, Turner, Williams & Haselkorn, 2011), women with a diagnosis of MS (Beal & Stuifbergen, 2007; Harrison & Stuifbergen, 2002), individuals with a diagnosis of relapsing-remitting MS specifically (Dlugonski & Motl, 2012; Effat et al. 2016; Suh, Weikert, Dlugonski, Sandroff & Motl, 2012), mothers with a diagnosis of MS (Gulick & Kim, 2004), individuals with a diagnosis of MS who were hospitalised on a neurological ward (Jaracz et al., 2010) and non-hospitalised individuals with a diagnosis of spinal cord form of MS (McIvor et al., 1984). Of the four studies that included comparison groups (Bamer, Cetin, Johnson, Gibbons & Ehde, 2008; Effat et al., 2016; Jaracz et al., 2010; McCabe et al., 2004), one used an 'eastern' and 'western' sample from the USA (Bamer et al., 2008) and both samples included individuals

with a diagnosis of MS and were used to compare locations (urban and rural). The other three studies included an MS group and a control group with ‘healthy’ individuals for comparison (Effat et al., 2016; Jaracz et al., 2010; McCabe et al., 2004). Hence, a variety of general and specific MS populations are included in the review, with the majority (57%) being conducted with a general MS population, which included the varying types.

Of the studies for which a response rate was applicable ($n = 19$), 26% ($n = 5$) did not report a response rate (Costa et al., 2012; Effat et al., 2016; Gay et al., 2010; Jaracz et al., 2010; McIvor et al., 1984). Of the studies that did report a response rate the lowest response rate for a MS sample was Bamer et al., (2008) with a response rate of 39.3% in the ‘eastern sample’ group. The highest response rate for an MS sample was Hyarat et al. (2018) with a 100% response rate. There was only one study (Bambara et al., 2010) who reported the characteristics of non-responders to identify any potential responder bias.

The number of MS participants across all the studies totalled 4,628. This total was calculated following removal of the ‘healthy’ control group numbers ($n = 429$) and removal of duplicate samples (see Appendix 1-E for further information).

Demographic data for the participants with a diagnosis of MS were reported for 99% of the participants ($n = 4,599$). The demographic data for one study (Schwartz & Frohner, 2005) were not included in these totals as the authors provided demographic data for 82 participants but only included 69 participants in the analysis. Of the demographic data of the 4,599 participants reported, 73% were female ($n = 3,339$) which is in line with the prevalence rates of individuals with a diagnosis of MS (around 2-3 times more common in females than males; Raffel, Wakerley & Nicolas, 2016). For the ‘healthy’ control groups ($n = 429$), 65% were female ($n = 279$). For the participants with a diagnosis of MS and for whom the age range was reported ($n = 13$), the ages ranged from 18 to 95 years and for the ‘healthy’ control groups of which the age range was reported ($n = 2$), the ages ranged from 18 to 65 years.

Eleven of the studies reported ethnicity of the MS participants, and this information can be viewed in Table 2. There were no data on ethnicity for any of the comparison group studies.

Most of the studies ($n = 15$) had sample sizes of 100 or above, with four studies having a sample size lower than 75 (Dlugonski & Motl, 2012; Farran et al., 2016; Foote et al., 1990; Schwartz & Frohner, 2005). One study conducted by Effat et al. (2016) had a total of 90 participants but only 60 of these were individuals with a diagnosis of MS, with the other 30 participants in a comparison group. The study with the largest sample size was Bamer et al. (2008) which had a sample size of 1,171 participants.

Insert Table 2 here please

Measures

The most commonly used social support measure was the Medical Outcomes Study Modified Social Support Scale (MSSS; Sherbourne & Stewart, 1991) which was used in six of the studies. All the other measures of social support were used less frequently and can be seen in Table 2. The most frequently measured form of psychological distress within the studies was depression ($n = 10$) and the most widely used measures of depression were the Patient Health Questionnaire (PHQ-9; Kroenke, Spitzer & Williams, 2001), the Center for Epidemiologic Studies Depression Scale (CES-D; Radloff, 1977) and the Beck Depression Inventory (BDI; Beck, Ward, Mendelson, Mock & Erbaugh, 1961), all of which were used twice. The measures used in each study, including their reliability and validity, were reviewed during the quality assessment process. All the studies scored a + for this area, indicating that the studies may not have addressed all potential sources of bias for this aspect

of study design, as all the measures used were subjective self-report measures. Although this is controversial by criticising self-report, this is how the quality assessment tool rated self-report measures. Moreover, the reliability and validity of the measures was either mentioned in the articles or were checked by the author.

McCabe et al. (2004) used four subscales (depression, tension, fatigue and confusion) of the POMS-SF (the Profile of Moods States – Short Form; Shacham, 1983) to measure adjustment. They defined adjustment as depression, anger, anxiety, fatigue and confusion and they used the tension subscale to measure anger and anxiety. For the purposes of this review the confusion and fatigue components were not required, therefore only the scores for depression and tension components were used. Hence, these scores were not conceptualised as adjustment, rather the individual components of depression, anger and anxiety.

Statistical Analyses

The statistical analyses employed across the studies were considered appropriate for the study designs; five of the studies conducted correlational analyses only, a further four studies conducted different types of regression analyses and the remaining 12 studies combined both correlational and regression analyses. Two of the 12 studies which combined correlational and regression analyses (McIvor et al., 1984; O'Brien, 1993) did not report the variance contributed by social support to the model separately and therefore the results of these could not be reported.

Findings

Indirect pathway: individual coping resources.

The following results depict an indirect effect of social support on outcomes of psychological distress using individual coping resources, based on the theoretical framework of Heaney and Israel (2008). This suggests that social support can enhance an individual's ability to problem solve and cope with health-related stressors which then influences their

physical, mental and social health. Coping has been defined as a form of behaviour which aims to manage internal and environmental demands that exhaust an individual's resources (Lazarus & Folkman, 1984). Coping can be separated into two forms; problem-focused and emotion-focused. Problem-focused coping relates to efforts aimed to manage the source of stress directly by altering the situation, whereas emotion-focused coping aims to manage the source of stress by dealing with the emotional reaction (O'Brien, 1993).

Coping.

One study investigated the relationship between social support and problem-focused coping behaviour (O'Brien, 1993). No statistically significant correlation was identified between social support and problem-focused coping.

Direct pathway.

The following results depict a direct effect of social support on outcomes of psychological distress based on the theoretical framework of Heaney and Israel (2008). This suggests that social support is directly related to physical, mental and social health which then influences health behaviours.

Anger and anxiety.

One study (McCabe et al., 2004) investigated the relationship between social support, anger and anxiety, measured using the tension component of the POMS-SF. More social support was a significant predictor of less anger and anxiety for female participants, but not males.

Anxiety.

Koelmel et al., 2017 studied the relationship between social support and anxiety using a longitudinal design. Statistically significant, although small, negative correlations were found between social support and anxiety for at least one of the social support components at each of the time points, with higher levels of social support associated with

lower levels of anxiety. They also identified that greater social support was moderately predictive of less anxiety at subsequent time points.

Depression.

Ten studies assessed the relationship between social support and depression, of these, seven conducted correlational analysis, which all found statistically significant negative correlations, where greater social support was associated with less depression. These significant results ranged from small effect sizes, above .10, (Bambara et al., 2011), medium effect sizes, above .30 (Gay et al., 2010; Harrison & Stuifbergen, 2002; Suh et al., 2012) to large effect sizes, above .50 (Hyarat et al., 2018; McIvor et al., 1984). Koelmel et al. (2017) conducted a longitudinal study and found statistically significant, although small, negative correlations for a least one of the social support components at each of the time points, with greater social support being associated with less depression.

These significant relationships mainly remained in the regressions, greater social support was a significant predictor of less depression over and above demographic and clinical variables (Bambara et al., 2011; Bamer et al., 2008; Chwastiak et al., 2002). McCabe (2004) also identified more social support as a significant predictor of less depression for female participants. However, the same relationship was not identified for male participants. Harrison and Stuifbergen (2002) also identified more social support as a significant predictor for less depression in mothers with MS, over and above the other variable included in the model, concern for children. Suh et al. (2012) also found that more social support was a significant predictor of less depression when included with physical activity and social support remained significant even when mobility disability and perceived stress entered the model. Koelmel et al.'s, (2017) longitudinal study identified that greater social support was moderately predictive of less depression at subsequent time points.

Hope.

One study (Foote et al., 1990) found a statistically significant positive correlation, with a large effect size, between social support and hope, with higher levels of social support associated with higher levels of hope.

Loneliness.

One study (Beal & Stuifbergen, 2007) assessed the relationship between social support and loneliness; a correlational analysis showed higher levels of social support were associated with lower levels of loneliness, with medium effect size. This relationship remained in the regression, as more social support was found to be a significant predictor of less loneliness, over and above the variables of functional limitation, self-rated health status, social response of illness and marriage.

Mental aspects of health-related quality of life.

Five studies investigated the relationship between social support and mental health aspects of quality of life. Of these, four conducted correlational analyses with three of these finding significant positive correlations for all aspects of social support components (Farran et al., 2016; Schwartz & Frohner, 2005; Jaracz et al., 2010), with greater social support being associated with better mental aspects of quality of life. Of these, two reported large effect sizes (Farran et al., 2016; Schwartz & Frohner, 2005) and Jaracz et al. (2010) reported a mix of small and medium effect sizes for the different components of social support. Effat et al. (2016) reported mixed results. They found statistically significant positive correlations between social support and the vitality and mental health components of the MSQLI, with medium effect sizes. Higher levels of social support were associated with better mental-health related quality of life. Conversely, no significant correlations were identified between social support and the social functioning and role emotion components, in contrast to the results found by Schwartz & Frohner (2005) who used the same measure of mental health aspects of quality of life.

Results were also mixed at regression, as Costa et al. (2012) found that only the psychological support component of social support was a significant predictor of all aspects of the mental components of health-related quality of life when demographic, clinical and disability variables were included in the model. The material support component was not identified as a significant predictor for any of the mental components of health-related quality of life. Jaracz et al. (2010) found that social support was not identified as a significant predictor when included with demographic, clinical and depression variables in the model. However, when depression was removed from the model, social support became a significant predictor, over and above demographic and clinical variables. Schwartz and Frohner (2005) found a similar result, as they only included demographic and clinical factors alongside social support and found social support to be a significant predictor of mental health related quality of life beyond all other variables.

Mental health status.

One study (Koelmel et al., 2017) investigated the relationship between social support and general mental health status longitudinally. Statistically significant positive correlations, ranging from small to medium effect sizes, were found between social support and general mental health status for at least one of the social support components at each time point. Greater social support was associated with better general mental health status. These significant findings remained at regression as they identified that greater social support was moderately predictive of better general mental health status at subsequent time points.

Postpartum emotional distress.

One study (Gulick & Kim, 2004) assessed the relationship between social support and postpartum emotional distress, using longitudinal correlational and regression analyses. They found that all, but one, deficit components of social support (difference between support received and importance of this support) showed statistically significant positive correlations

with emotional distress at each of the time points, with greater support deficit associated with more emotional distress. The only exception was for the instrumental support deficit component at month one, as a statistically significant negative correlation was identified, suggesting that the greater the instrumental support deficit, the less emotional distress. Interestingly, this relationship reversed at three months and six months as a statistically significant positive correlation was identified. The effect sizes of these correlations ranged from small to large. Regressions were conducted at the three time points and only the emotional support deficit component entered the model. Greater emotional support deficit was identified as a significant predictor of increased postpartum emotional distress at all three timepoints.

Psychosocial adjustment.

One study (Sullivan et al., 2004) assessed the relationship between social support and psychological adjustment, which they conceptualised as healthcare orientation, vocational environment, domestic environment, sexual relationships, social environment and psychological distress (measured using a single scale: Psychosocial Adjustment to Illness Scale; PAIS; Derogatis, 1986). They found a statistically significant, although small, negative correlation between social support and psychological adjustment, with higher levels of social support associated with better adjustment. They also found that social support was not a statistically significant predictor of adjustment in the regression.

Self-esteem.

Two studies (Dlugonski & Motl, 2012; Foote et al., 1990) investigated the relationship between social support and self-esteem, using both correlational and regression analyses. Statistically significant positive correlations were found between social support and self-esteem, with medium effect sizes, whereby higher social support was associated with higher self-esteem. This finding remained at regression as increased social support was

found to be a statistically significant predictor of increased self-esteem, over and above physical activity.

Quality Assessment

Only one of the 21 articles reviewed scored the maximum score for both external and internal validity (Schwartz & Frohner, 2005). There was only one study which received a single negative rating for either of the validity components (Foote et al., 1990) which was for internal validity. All the other studies scored at least one positive rating or the highest rating for either or both components. This suggests that the overall quality of the papers can be considered as moderate to good results.

Only one study (O'Brien, 1993) failed to find a statistically significant correlation, which was between social support and coping. This study had not reported a power calculation but had a sample size of 101 participants, sufficient to demonstrate a medium effect. It scored a + rating on the quality assessment due to the sample size and the representation of the sample was similar to that of the prevalence of MS (gender ratio and ethnicity). One aspect in which there was a lack of information was the question regarding the completion of the outcome measurements, as only six studies reported information regarding missing data and completion rates of the outcome measures. All the studies which reported this scored positively on this question (Bambara et al., 2011; Bamer et al., 2008; Dlugonski & Motl, 2012; Foote et al., 1990; Koelmel et al., 2017; Sullivan et al., 2004). As the other fifteen studies did not report this information, it is not possible to determine whether there were any missing data and if so, how this had been managed.

All the studies used a cross-sectional design, with only two studies also including a longitudinal element (Gulick & Kim, 2004; Koelmel et al., 2017). A potential limitation to using this design is the lack of evidence surrounding causality; cross-sectional study designs do not allow for a distinction between cause and effect (Mann, 2003).

The most positively reported items on the assessment was the section relating to analyses, with seven studies (Bambara et al., 2011; Bamer et al., 2008; Chwastiak et al., 2002; Costa et al., 2012; Gulick & Kim, 2004; Jaracz et al., 2010; Koelmel et al., 2017) scoring the maximum rating of ++ for each question in the section.

Discussion

Each paper, except one (O'Brien, 1993), included in this systematic review reported at least one statistically significant finding in relation to perceived social support and psychological variables in individuals with a diagnosis of MS. The results found were consistent with the direct pathway of the theoretical model proposed by Heaney and Israel (2008) as positive correlations were found between social support and these variables (hope, mental aspects of health-related quality of life, mental health status, postpartum emotional well-being and self-esteem), whereby higher levels of social support were associated with higher levels of these variables. Social support was also found to negatively correlate with anxiety, anger, depression, loneliness, psychological adjustment, where greater social support was associated with lower levels of these variables. In regressions, greater social support was shown to be a significant predictor of less anxiety, anger, depression and loneliness. Furthermore, greater social support was also found to be a significant predictor of better mental health aspects of health-related quality of life, postpartum emotional well-being and self-esteem. These findings were still significant when other demographic, clinical and psychological variables were included in the model.

Conversely, the results were not consistent with the indirect pathway through individual coping resources proposed by Heaney and Israel (2008), as non-significant correlations were identified for the only variable included in this pathway: problem-focused coping (O'Brien, 1993). Non-significant findings were also reported in two studies at regression, these were for psychosocial adjustment (Sullivan et al., 2004), and for depression

and tension (anger and anxiety) in male participants (McCabe et al., 2004) which were included in the direct pathway.

Some of these findings were stronger than others, as some of these relationships were only identified in one or two studies (anxiety, anger, loneliness, hope, mental health status, postpartum, psychological adjustment and self-esteem). However, the relationship with other variables such as depression and mental aspects of health-related quality of life were identified in multiple studies, with depression being studied the most frequently. Furthermore, various effect sizes were reported, with the largest effect sizes identified for studies that assessed depression, hope, mental aspects of health-related quality of life and postpartum emotional distress. However, not all the studies assessing depression and mental aspects of health-related quality of life identified large effect sizes, as some reported small and medium effect sizes and one study (McCabe et al. 2004) reported non-significant results for male participants at regression. Due to the longitudinal aspects of the study assessing postpartum emotional distress (Gulick & Kim, 2004), the effect sizes also varied at the different time points and included small and medium effect sizes too. Moreover, those studies using a correlational design only, have less strength than those reporting regressions. Therefore, when considering all the aspects relating to the strength of these findings, depression followed by mental aspects of health-related quality of life appear to be the strongest. These had large effect sizes at correlation, were also found to be significantly predicted by social support at regression and were assessed by multiple studies. Mental aspects of health-related quality of life and depression also had some of the highest quality assessment ratings.

The current findings are similar to those found by Dennison et al. (2009) whose definition of adjustment included psychological well-being; therefore, variables that this review considered as evidence of 'adjustment' were also included here. This included

variables such as anger, anxiety, depression and mental aspects of health-related quality of life. Social support was identified as a significant predictor of these variables, as they were in the Dennison et al. (2009) review. The present review builds on this, incorporating more up to date research as only three studies in the present review overlapped with theirs (McCabe et al., 2004; McIvor et al., 1984; Schwartz & Frohner, 2005).

Depression is another variable included in the current review that previous research has identified as a correlate of social support. A review of depression in MS (Arnett et al. 2008) identified a consistent relationship between social support and depression, therefore the results of this review are congruent with and update these findings. Only two of the eight depression studies included in the current review (Chwastiak et al., 2002; McIvor et al., 1984) were included in the Arnett et al, (2008) review.

These findings are congruent with other theoretical perspectives of social support, such as the stress, social support, and the buffering hypothesis, proposed by Cohen and Wills (1985). This puts forward two models to account for the process through which social support has a positive impact on well-being in the context of physical health conditions; the main effects and stress buffer models. The main effects model suggests that social support could be beneficial to psychological and health outcomes due to the individual regularly receiving positive experiences and gaining socially rewarded roles in the community. This can in turn promote positive affect, predictability, stability and an enhancement of self-worth. The stress buffer model suggests that stress can be experienced as a result of illness, failures in self-care and by disruptions to the neuroendocrine or immune system functioning. This stress can contribute to negative affect, increase in physiological responses, behavioural adaptations, feelings of helplessness and reductions in self-esteem. This model suggests that social support can influence the negative impacts of stress at two points in the causal chain between stress and illness. Social support could mediate between the stressful (or anticipated

stressful) event and the stress reaction by preventing a stress appraisal response.

Alternatively, sufficient support could mediate between the experience of stress and the resulting physiological response by reducing the stress reaction or directly affecting the physiological processes. The findings from this review are congruent with the main effects model, however, more research is needed to explore the mechanisms through which this relationship is produced. Further research using more complex models would be beneficial to explore the complex relationship that social support has on improving psychological distress and enhancing well-being for individuals experiencing physical health conditions, including MS.

Strengths and Limitations of the Current Review

Although the majority of studies ($n = 13$) were conducted in the USA, this review was able to include studies from several countries, which increases generalisability. Another strength of this review is in terms of the populations used within the studies, as a good range of populations were included, such as; hospitalised, non-hospitalised, spinal form MS, RRMS, mothers, women and veterans.

One limitation regards the diagnosis of MS, as no exclusion criteria was set for this. Nine of the studies (43%) used self-report to determine this which could allow for individuals who may not have a definite diagnosis of MS to participate, potentially reducing the validity of the sample.

A further consideration regards the measure used to conceptualise loneliness in the Beal and Stuifbergen (2007) study. To measure loneliness, they used one question from the CES-D "I felt lonely", although this is not a specific measure of loneliness, making this less psychometrically robust. However, this question was taken from a reliable and valid measure and this question had been designed to measure loneliness as a part of this measure. A similar question of loneliness was found to highly correlate with the Revised UCLA

Loneliness Scale (Russell, Peplau & Cutrona, 1980) which is a widely used measure of loneliness in research.

Clinical Implications

These findings highlight the important role that social support can have in predicting psychological outcomes in individuals with MS. The outcome with the most prominent relationship with social support was depression, and research has estimated lifetime prevalence rates of major depression of up to 50% for individuals with MS (Feinstein, 2011). It would therefore be useful for clinicians working with individuals with MS who are experiencing psychological distress, such as depression, to establish an individuals' levels of perceived social support when assessing their psychological needs. Social support interventions, such as peer support, have been found to be beneficial in reducing levels of depression in mental health populations (see meta-analysis by Pfeiffer, Heisler, Piette, Rogers & Valenstein, 2011).

Research investigating the effectiveness of social support interventions for individuals with MS is limited. One review of social support interventions in a variety of populations was conducted by Hogan, Linden and Najarian (2002), who identified some findings that supported their use. This included research conducted by Maton (1988), which included participants who had attended a MS peer support group and found that those providing support and those who both provided and received support reported higher levels of well-being. Ng, Amatya, and Khan (2013) evaluated the impact of a peer support programme on improving MS psychological functioning (depression, anxiety and stress). Participants reported improved psychological functioning and quality of life six weeks following the programme, and positive improvements in stress and quality of life were maintained at 12 months follow up. However, Uccelli, Mohr, Battaglia, Zagami and Mohr (2004) assessed the effectiveness of peer support groups in MS and found that attendance at support groups did

not improve depression or quality of life. Moreover, those who had better mental health functioning could be at risk of deterioration by accessing the group. Thus, further research is required to understand the best way of offering social support, as the current literature is unclear.

Future Research

Some variables included in this review were only investigated by one study (anger, coping, hope, loneliness, mental health status, postpartum emotional distress and psychological adjustment) so further research in these areas would help to enhance the strength of the relationships found. Anxiety was also studied infrequently, and this is especially important as anxiety is common in MS but is often over-looked and under-treated (Korostil & Feinstein, 2007). Anger has also been identified as an important psychological variable for individuals with a diagnosis of MS, as levels of anger have been found to be higher or lower than in the general population (Nocentini et al., 2009). These levels of anger were found to be mainly independent of mood and the authors suggest this may have a physiological cause. Therefore, further exploration of these variables would benefit the understanding and resulting management of these difficulties.

Only three of the studies included in this review (Bambara et al., 2011; Chwastiak et al., 2002; Sullivan et al., 2004) assessed the differences between outcome measure scores and the type of MS. Chwastiak et al. (2002) identified no significant difference between the mean depression scores of three subtypes of MS (relapsing-remitting, primary progressive and secondary progressive). However, Bambara et al. (2011) found that those with a diagnosis of progressive MS had higher levels of depression than those with a relapsing-remitting subtype. This was supported by Sullivan et al. (2004) as they identified that those in remission had significantly better levels of adjustment than those in exacerbation or with the secondary-progressive subtype (there was no difference identified between those in

exacerbation and those with secondary-progressive subtype). As these studies highlight, some significant differences between the subtypes of MS can occur, and by including and comparing the different subtypes within the samples and including these within regression analyses, strengthen their findings. Including this comparison within future research would help to establish whether a certain subtype has a stronger relationship to psychological distress than another.

Furthermore, investigating the mechanisms of how social support influences well-being would also be beneficial to understand how better to support people once difficulties have been identified. More longitudinal studies are required as only two studies using a longitudinal design were identified in this review (Gulick & Kim, 2004; Koelmel et al., 2017). More complex models such as those conducted by Koelmel et al., (2017) whereby the effect that social support had on mental health outcomes at subsequent time-points by using a linear mixed-effects regression analyses would enhance future findings.

Conclusion

The results of this systematic literature review provide evidence for some significant relationships between perceived social support and anxiety, anger, depression, hope, loneliness, mental aspects of health-related quality of life, mental health outcomes, postpartum emotional distress, self-esteem and psychological adjustment in individuals with a diagnosis of MS. However, most of the research is cross-sectional, many studies are only correlational and limited research exists on certain variables. Useful implications for clinical practice, such as highlighting the relationship between social support and psychological outcomes and facilitating access and receipt of this could enhance individual's psychological well-being.

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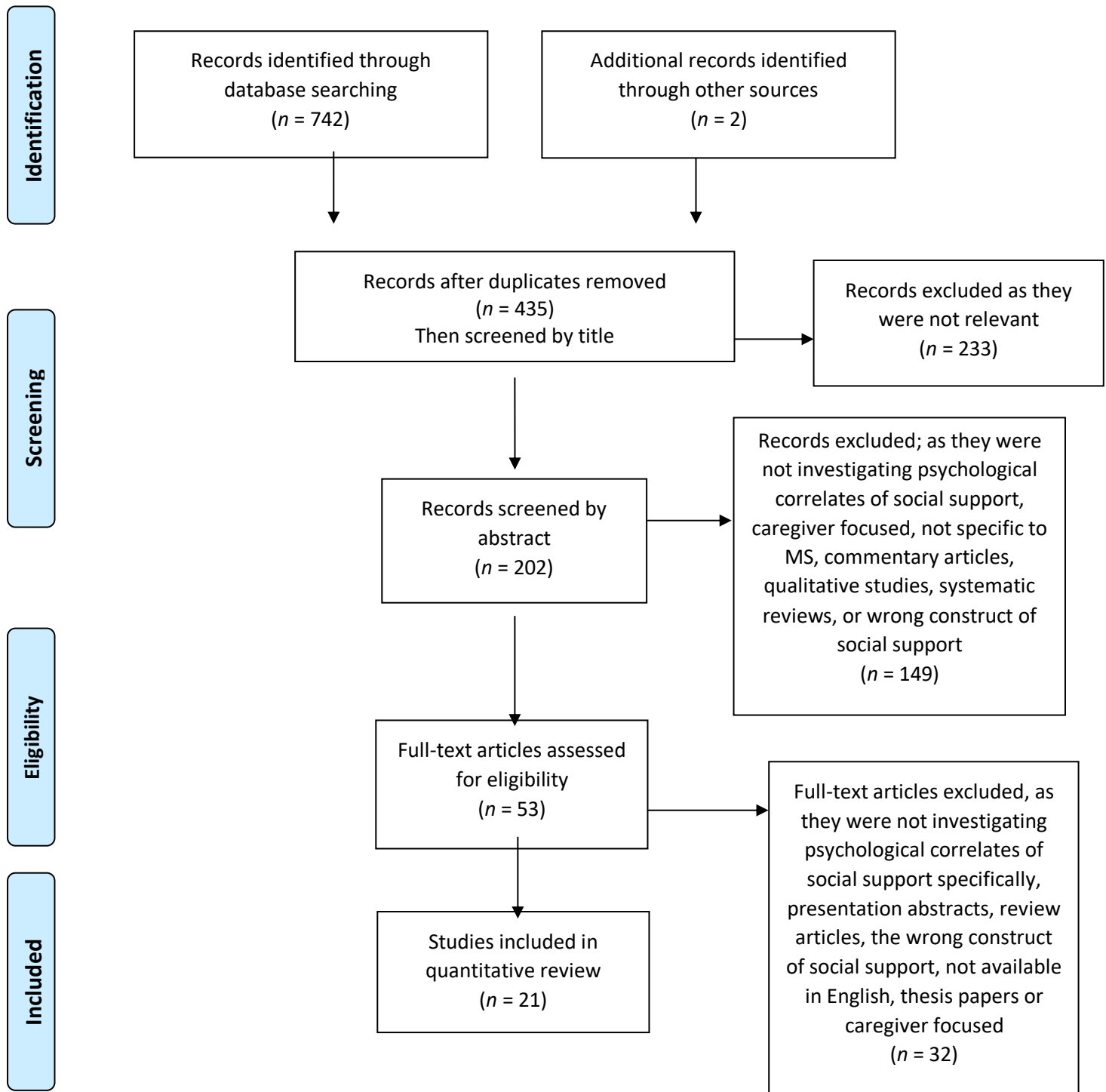
Figure 1: Flow diagram showing the process of identifying articles (PRISMA, 2009)

Table 1: *Quality assessment ratings (NICE, 2018)*

Study	1.1	1.2	1.3	2.1	2.2	2.3	2.4	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.6	Total Internal Validity	Total External Validity
Bambara et al. (2011)	+	+	+	NA	++	NA	++	+	++	NA	NA	NA	++	++	++	++	++	+
Bamer et al. (2008)	++	+	+	++	+	NA	++	+	++	NA	NA	NA	++	++	++	++	++	+
Beal and Stuifbergen (2007)	+	+	+	NA	++	NA	+	+	NR	NA	NA	NA	++	+	+	++	+	+
Chwastiak et al. (2002)	+	+	+	NA	+	NA	++	+	NR	NA	NA	NA	++	++	++	++	++	+
Costa et al. (2012)	+	+	+	NA	++	NA	++	+	NR	NA	NA	NA	++	++	++	++	++	+
Dlugonski and Motl (2012)	+	+	+	NA	++	NA	+	+	++	NA	NA	NA	-	+	+	++	+	+
Effat et al. (2016)	+	+	+	NA	+	NA	-	+	NR	NA	NA	NA	-	++	-	++	+	+
Farran et al. (2016)	++	++	+	NA	+	NA	-	+	NR	NA	NA	NA	-	++	-	++	+	++
Foote et al. (1990)	+	++	++	NA	++	NA	-	+	++	NA	NA	NA	-	-	-	++	-	++
Gay et al. (2010)	+	+	+	NA	++	NA	-	+	NR	NA	NA	NA	+	++	-	++	+	+
Gulick and Kim (2004)	+	++	+	NA	++	NA	++	+	NR	NA	NA	+	++	++	++	++	++	+
Harrison and Stuifbergen (2002)	+	+	+	NA	++	NA	+	+	NR	NA	NA	NA	++	+	+	++	+	+

Study	1.1	1.2	1.3	2.1	2.2	2.3	2.4	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.6	Total Internal Validity	Total External Validity
Hyarat et al. (2018)	+	++	+	NA	++	NA	-	+	NR	NA	NA	NA	++	+	-	++	+	+
Jaracz et al. (2010)	++	+	+	++	++	NA	++	+	NR	NA	NA	NA	++	++	++	++	++	+
Koelmel et al. (2017)	+	+	+	++	+	++	++	+	+	NR	++	++	++	++	++	++	++	+
McCabe et al. (2004)	+	+	+	++	++	NA	++	+	NR	NA	NA	NA	++	++	++	NR	++	+
McIvor et al. (1984)	+	+	+	NA	++	NA	++	+	NR	NA	NA	NA	+	+	+	++	+	+
O'Brien (1993)	+	+	+	NA	++	NA	-	+	NR	NA	NA	NA	+	+	-	NR	+	+
Schwartz and Frohner (2005)	+	++	++	NA	++	NA	++	+	NR	NA	NA	NA	-	++	++	++	++	++
Suh et al. (2012)	+	+	+	NA	++	NA	+	+	NR	NA	NA	NA	++	+	+	++	+	+
Sullivan et al. (2004)	+	+	+	NA	+	NA	++	+	++	NA	NA	NA	+	++	++	+	++	+

Questions: The NICE quality appraisal checklist – quantitative studies reporting correlations and associations (p. 200 - 205; 2018)

Section 1: Population

1.1 - Is the source population or source area well described?

1.2 - Is the eligible population or area representative of the source population?

1.3 - Do the selected participants or areas represent the eligible population or area?

Section 2: Method of selection of exposure (or comparison) group

- 2.1 - Selection of exposure (and comparison) group. How was selection bias minimised?
- 2.2 - Was the selection of explanatory variables based on a sound theoretical basis?
- 2.3 - Was the contamination acceptably low?
- 2.4 - How well were likely confounding factors identified and controlled?
- 2.5 - Is the setting applicable to the UK?

Section 3: Outcomes

- 3.1 - Were the outcome measures and procedures reliable?
- 3.2 - Were the outcome measurements complete?
- 3.3 - Were all important outcomes assessed?
- 3.4 - Was there a similar follow-up time in exposure and comparison groups?
- 3.5 - Was follow-up time meaningful?

Section 4: Analyses

- 4.1 - Was the study sufficiently powered to detect an intervention effect (if one exists)?
- 4.2 - Were multiple explanatory variables considered in the analyses?
- 4.3 - Were the analytical methods appropriate?
- 4.6 - Was the precision of association given or calculable? Is association meaningful?

Section 5: Summary

- 5.1 - Are the study results internally valid (i.e. unbiased)? – **Total Internal Validity**
- 5.2 - Are the findings generalisable to the source population (i.e. externally valid)? – **Total External Validity**

Scale

++ indicates that for that particular aspect of study design, the study has been designed or conducted in such a way to minimise the risk of bias.

+ indicates that either the answer to the checklist question is not clear from the way the study is reported, or that the study may not have addressed all potential sources of bias for that particular aspect of study design.

- should be reserved for those aspects of the study design in which significant sources of bias may persist.

NR not reported should be reserved for those aspects in which the study under review fails to report how they have (or might have) been considered.

NA not applicable should be reserved for those study design aspects that are not applicable given the study design under review.

Table 2: *Articles included in the review*

Study	Participants	Recruitment	Origin	Outcome Measures	Response Rates	Correlate	Other variables included in regression	Type of Analysis	Results
Bambara et al. (2011)	451 veterans with diagnosis of MS (390 males; mean age 55.1; 415 Caucasian)	Northwest Regional database of Veterans Health Administration	USA	MSSS M-PS PHQ-9	44% response rate	Depression	Demographic and clinical variables	Exploratory correlation analyses and hierarchical multiple regression	<p>Correlations: Emotional/informational support, affection, positive social interactions and total social support were significantly related to depression ($r = -0.20, p < 0.001$; $r = -0.14, p < 0.001$; $r = 0.20, p < 0.001$, $r = -0.17, p < 0.001$ respectively)</p> <p>Regression: greater perceived social support was significantly associated with less depression after accounting for demographic variables, MS disease-related</p>

									variables and living situation ($\beta = -0.24, p < 0.001$)
Bamer et al. (2008)*	<p>1171 participants with a diagnosis of MS (661 western sample and 510 eastern sample)</p> <p>Western Sample: 364 relapsing-remitting, 126 primary progressive, 210 secondary progressive; mean duration of 12.5 years; 576 females; mean age 49.2; 674 Caucasian</p> <p>Eastern Sample: 262 relapsing-remitting, 47 primary</p>	<p>Western Sample: Survey sent to members of the Multiple Sclerosis Association of King County</p> <p>Eastern Sample: Survey sent to members of the National Multiple Sclerosis Society Inland Northwest Chapter</p>	USA	MSSS EDSS CES-D	<p>Western Sample: 50.8% response rate</p> <p>Eastern Sample: 39.3% response rate</p>	Depression	Demographic and clinical variables	Multivariate logistic regression	<p>Western Sample: A lack of social support was significantly associated with higher levels of depression on the CES-D ($\chi^2 = 54.91, p < 0.001$; Odds Ratio = 1.99; 95% CI = 1.66-2.39)</p> <p>Eastern Sample: A lack of social support was significantly associated with higher levels of depression on the CES-D ($\chi^2 = 30.36, p < 0.001$; Odds Ratio = 1.85; 95% CI = 1.49-2.31)</p>

	progressive, 169 secondary progressive; mean duration of 15 years; 398 females; mean age 52.4; 502 Caucasian								
Beal and Stuifbergen (2007) **	659 females with a diagnosis of MS (316 relapsing-remitting, 217 chronic progressive, 126 not reported; average duration of 10 years, range 1-46; 659 females, mean age 47, range 18-95; ethnicity: 606 White, 26 African American, 17 Hispanic, 10 not reported)	Secondary analysis of baseline data collected for a longitudinal investigation. Contacted through two southwestern USA chapters of the National Multiple Sclerosis Society ⁷	USA	PRQ Part 2 CES-D ISS MAI DOII	88.8% response rate for initial investigation ***	Loneliness	Functional limitation, self-rated health status, social response of illness and marriage status	Pearson correlation and stepwise regression analysis	<p>Correlation: A moderate negative relationship was found between social support and loneliness ($r = -0.45, p < 0.01$)</p> <p>Regression: Social support was found to be a significant predictor of loneliness ($\beta = -0.34, p < 0.05$)</p>

Chwastiak et al. (2002) *	739 participants with a diagnosis of MS (course of MS reported [$n = 714$] 369 relapsing-remitting, 130 primary progressive, 215 secondary progressive; mean duration of 12.5 years; 575 females; mean age 49.3, range 21-83; 675 Caucasian)	Survey sent to members of the Multiple Sclerosis Association of King County (Washington)	USA	CES-D EDSS MSSS	53.8% response rate	Depression	Demographic and clinical variables	Multivariate logistic regression modelling	Lack of social support was significantly associated with higher levels of depression on the CES-D ($\chi^2 = 49.5$, $p = 0.001$; Odds Ratio = 1.92; 95% CI = 1.60-2.30)
Costa et al. (2012)	150 patients with diagnosis of definite MS (128 relapse-remitting, 6 primary progressive, 16 secondary progressive; median duration 9.1	Consecutive patients attending MS Clinic of Hospital S. João, Porto	Portugal	MSSS SF-36v2 EDSS	Not reported	Mental dimension of health-related quality of life	Demographic, clinical and disability variables	Multiple linear regression	The psychological support component of the MSSS produced a statistically significant change in the vitality, social functioning, emotional performance and mental health components of the

	years; 106 females, median age 41.7, range 18-70)								mental health dimension of the SF-36v2 ($\beta = 0.290, p = 0.000$; $\beta = 0.336, p = 0.000$; $\beta = 0.214, p = 0.003$; $\beta = 0.447, p = 0.000$ respectively). No statistically significant changes were reported for the material support component of the MSSS
Dlugonski and Motl (2012)	46 participants with a diagnosis of relapsing-remitting MS (average duration of 9.2 years; 40 females; average age 46.5; 44 Caucasian)	Convenience sample from a large database of individuals with MS who were interested in research opportunities from one laboratory. Recruitment was conducted via email.	USA	SPS RSES MSIS-29 PDDS	83.6% response rate ***	Self-esteem	Physical activity	Bivariate correlation and multiple linear regression	<p>Correlation: A statistically significant correlation was found between social support and self-esteem ($r = 0.366, p < 0.01$)</p> <p>Regression: Social support was a statistically significant predictor of self-esteem ($\beta = 0.411, p = 0.004$)</p>
Effat et al. (2016)	MS Group: 60 participants	Recruited from outpatient clinics of two	Egypt	MSSS EDSS BDI	Not reported	Mental dimension of health-	Not applicable	Pearson correlation coefficient	Social support significantly correlated with

	with a definite diagnosis of relapse-remitting MS (mean duration of 4.14 years; 32 females; mean age 35.47, range 22-44) Comparison Group: 30 healthy controls (15 females; mean age 34.1)	hospitals in Egypt		MSQLI		related quality of life			two of the mental components (vitality and mental health) of the MSQLI ($r = 0.333, p < 0.01$; $r = 0.437, p < 0.001$ respectively)
Farran et al. (2016)	34 participants with self-reported physician confirmed diagnosis of MS (22 relapsing-remitting, 5 patient did not know, 4 secondary progressive, 2 primary progressive,	Recruited via social media within Lebanon, including online groups for people with MS.	Lebanon	SPS MusQoL BDI-II BAI FSS	Not applicable (online recruitment)	Mental health composite of quality of life (MHQOL)	Not applicable	Pearson correlation coefficient	Social support was positively correlated with QoL ($r = 0.728, p < 0.001$)

	1 progressive relapsing; 19 females, mean age 36)								
Foote et al. (1990)	40 participants with a diagnosis of MS (32 female; mean age 48.2, range 32-70; ethnicity: 37 Caucasian, 3 Non-Caucasian)	Convenience sample of contacted through a large south-eastern MS Clinic	USA	PRQ part 2 MHS RSES	57.1% response rate ***	Hope and self-esteem	Not applicable	Pearson correlation coefficient	Social support was statistically significantly correlated with both hope and self-esteem ($r = 0.6848, p < 0.001$; $r = 0.4352, p < 0.01$)
Gay et al. (2010)	115 participants with MS (79 females; mean age 47.22, range 27-80; 101Caucasia n)	Recruited through various associations, MS departments in Parisian hospitals and private neurologists	France	SSQ-6 EDSS DSRS STAI SEI TAS 20 CHIP	Not reported	Depression	Not applicable	Pearson correlations	Correlation: Social support significantly correlated with depression ($r = -0.35, p < 0.01$)
Gulick and Kim (2004)	174 females who were mothers with a diagnosis of MS (average duration 5 years; average age	Convenience sample recruited via Web announcements in the Consortium of Multiple Sclerosis	USA	MSRS MSRSS PSQ	Not applicable (online and newsletter recruitment)	Postpartum emotional distress	Clinical variables and fatigue	Pearson correlation analyses and hierarchical linear regression analyses	Correlations: All deficit components of social support (emotional instrumental and informational) showed statistically significant

	32.7; ethnicity: 165 White, 4 Hispanic, 2 Black and not Hispanic, 2 American Indian or Alaskan Native, 1 Asian or Pacific Islander	Centers North American Research Committee on MS							<p>correlations with emotional distress at 1 month postpartum ($r = 0.50, p < 0.01$; $r = -0.34, p < 0.01$; $r = 0.20, p < 0.01$ respectively); 3 months ($r = 0.43, p < 0.01$; $r = 0.34, p < 0.01$; $r = 0.21, p < 0.01$ respectively); 6 months ($r = 0.49, p < 0.01$; $r = 0.39, p < 0.01$; $r = 0.31, p < 0.01$ respectively)</p> <p>Regressions: The instrumental and informational deficit support components failed to enter the model. The emotional support deficit component was strongly correlated with emotional distress at all three timepoints ($\beta = 0.34, p < 0.001$; $\beta = 0.23, p < 0.01$; $\beta = 0.38, p < 0.001$)</p>
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									at 1, 3 and 6 months respectively)
Harrison and Stuifbergen (2002) **	807 female participants with a physician-diagnosed MS (97 relapsing-remitting, 35 benign sensory, 55 progressive, 9 severe progressive; mean duration 10.05 years, range 1-45; 807 females, mean age 43, range 22-74; ethnicity: 185 White, 8 African American, 7 Hispanic, 1 Other)	Secondary analysis of baseline data collected for a longitudinal investigation. Contacted through national MS Society chapters	USA	PRQ 85 ISS CCS CESD-10	40% response rate for initial investigation	Depression	Concern for children	Pearson correlation and hierarchical multiple regression	<p>Correlation: Social support was statistically significantly correlated with depression ($r = -0.49, p < 0.01$)</p> <p>Regression: Social support was found to be a significant predictor of depression ($\beta = -0.43, p < 0.001$)</p>
Hyarat et al. (2018)	140 participants with a diagnosis of MS affirmed by medical	Contacted through out-patient clinics in King Abdulaziz Medical City,	Saudi Arabia	MSPSS BDI	100% response rate ***	Depression	Not applicable	Pearson correlation coefficient	A negative significant correlation was found between all aspects of the MSPSS (total

	records (range of duration 1-15 years; 99 males; mean age 34.3, range 18-59)	Ministry of National Guard Health Affairs in Riyadh.							score, family support subscale, significant other support subscale and friend support subscale) and depression scores ($r = -0.81, p < 0.005$; $r = -0.82, p < 0.005$, $r = -0.63, p < 0.005$ respectively)
Jaracz et al. (2010)	<p>MS Group: 210 patients with MS hospitalised in a neurological ward (mean duration 6.9 years, range 1-29; 150 females; mean age 37.4, range 21-59)</p> <p>Comparison Group: 108 healthy subjects (74 females; mean age 37.3 years, range 20-56)</p>	Recruited from a neurological ward	Poland	SPS MSQOL-54 BDI EDSS	Not reported	Mental health composite of quality of life (MHQOL)	Depression, demographic and clinical variables	Pearson correlation analysis and hierarchical regression analysis	<p>Correlations: All components of social support (guidance, reassurance of worth, social integration, attachment, nurturance, reliable alliance and total score) positively correlated with MHQOL ($r = 0.31, p < 0.01$; $r = 0.25, p < 0.01$; $r = 0.21, p < 0.01$; $r = 0.34, p < 0.01$; $r = 0.23, p < 0.01$; $r = 0.24, p < 0.01$; $r = 0.33, p < 0.01$ respectively)</p>

									Regressions: Social support was not identified as a significant predictor of MHQOL. However, once the variable of depression was removed, social support was a significant predictor of MHQOL ($\beta = 0.27, t < 0.001$)
Koelmel et al. (2017)	163 participants with a physician determined MS diagnosis (91 relapsing-remitting, 70 progressive, 2 not reported; mean duration 12 years, range 0-49; 142 females; mean age 52.2)	Accessed longitudinal data from a clinical trial, conducted through a university research registry and MS clinic, MS organisations, Clinical Trails website and other departmental studies	USA	MSPSS C-DRS PHQ-9 ED-A MCS	85.8% response rate ***	Mental health outcomes (depression, anxiety and general mental health status)	Time effects, demographic and clinical variables	Pearson product-moment correlations and linear mixed-effects regression analysis	Correlations: Statistically significant correlations were found between all types of social support (significant others, family members and friends) and depression at baseline ($r = -0.19, p < 0.05, r = -0.24, p < 0.01, r = -0.20, p < 0.05$ respectively). This was only partially supported at 26 weeks as support from

									<p>family members was no longer significant ($r = -0.19, p < 0.05, r = -0.09, r = -0.18, p < 0.05$ respectively) but stronger correlations were found at 52 weeks ($r = -0.23, p < 0.01, r = -0.19, p < 0.05, r = -0.20, p < 0.05$)</p> <p>Statistically significant correlations were found between family members and friends types of social support and anxiety at baseline ($r = -0.17, p < 0.05, r = -0.16, p < 0.05$ respectively) however at 26 and 52 weeks this changed to significant correlations found only between significant other component ($r = -0.24, p < 0.01, r =$</p>
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									<p>-0.20, $p < 0.05$ respectively). Statistically significant correlations were found between significant others and family members and general mental health status at baseline ($r = 0.18$, $p < 0.05$, $r = 0.23$, $p < 0.01$ respectively). All components were found to be statistically significant at 26 weeks ($r = 0.30$, $p < 0.001$, $r = 0.20$, $p < 0.05$, $r = 0.19$, $p < 0.05$ respectively). At 52 weeks, only significant others and family members were significant ($r = 0.25$, $p < 0.01$, $r = 0.17$, $p < 0.05$ respectively)</p> <p>Regressions: Each social support</p>
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									<p>subscale significantly predicted scores on general mental health outcomes. Greater social support predicted lower depression, lower anxiety and better general mental health status.</p> <p>Depression ($\beta = -0.18, p < 0.001, 95\% CI = -0.86$ to $-0.26; \beta = -0.23, p < 0.001, 95\% CI = -1.02$ to $-0.42; \beta = -0.20, p < 0.01, 95\% CI = -0.95$ to -0.29 for significant others, family members and friends respectively).</p> <p>Anxiety ($\beta = -0.16, p < 0.001, 95\% CI = -1.55$ to $-0.45; \beta = -0.19, p < 0.001, 95\% CI = -1.71$ to $-0.57; \beta = -0.17, p < 0.001, 95\% CI = -1.73$ to -0.52 for significant others,</p>
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									<p>family members and friends respectively).</p> <p>General mental health status ($\beta = 0.18, p < 0.001$, 95% <i>CI</i> = -0.07 to 0.19; $\beta = 0.18, p < 0.001$, 95% <i>CI</i> = 0.52 to 1.80; $\beta = 0.11, p = 0.03$, 95% <i>CI</i> = 0.07 to 1.47 for significant others, family members and friends respectively)</p>
McCabe et al. (2004)	<p>672 participants (381 with diagnosis of MS and 291 without MS)</p> <p>With MS: 237 females; mean age 44.45 years, range 18-65; 144 males, mean age 46.86, range 18-65</p>	<p>With MS: Randomly selected from the MS Society of Victoria, Australia register</p> <p>Without MS: Randomly selected from the electoral roll and screened for chronic illness</p>	Australia	SSF-WHOQOL-100 WOCQ POMS-SF (tension, depression, fatigue and confusion subscales)	<p>With MS: 60.5% response rate</p> <p>Without MS: 28.8% response rate</p>	Depression, anxiety and anger	Coping strategies and health related variables	Standard multiple regressions (with MS only)	<p>MS Females: Social support was a significant predictor of depression and tension components of the POMS-SF ($\beta = -0.16, p < 0.05$ $\beta = -0.19, p < 0.01$ respectively).</p> <p>MS Males: Social support was not found to be a significant predictor of</p>

	Without MS (general population): 190 females; mean age 45.35, range 18-65; 101 males; mean age 50.03, range 18-65								depression or anxiety and anger.
McIvor et al. (1984)	120 non-hospitalised patients with Spinal Cord form of MS (60 relapsing-remitting, 60 progressive; 88 females; mean age 45, range 23-71)	Consecutive patients under treatment at the Multiple Sclerosis Comprehensive Care Center, New York	USA	PSSI – Family-based and Friend-based WAIS (vocabulary subtest) BDI	Not reported	Depression	Not applicable	Pearson product-moment, biserial correlations and stepwise forward multiple regression analysis	Correlations: The correlations between social support (both family and friends) and depression were statistically significant ($r = -0.60, p < 0.001$; $r = -0.71, p < 0.001$ respectively) Regression: Results of individual variables not reported
O'Brien (1993)	101 participants with confirmed diagnosis of MS for at least one year (average	Convenience sample selected from local chapters of the National Multiple Sclerosis Society	USA	NSSQ WCC TSCS	71.6% response rate ***	Coping behaviour	Not applicable	Pearson product-moment correlation and multiple regression analysis	No statistically significant correlation was found between social support and problem-focused coping ($r = 0.126$)

	duration 12 years, range 1-41 years; 77 females; mean age 46.3, range 26-73; 81 White ethnicity)								Regression: Results of individual variables not reported.
Schwartz and Frohner (2005)	69 participants with a clinically definite diagnosis of MS for at least one year (demographic data not available)	Consecutive patients attending two clinics in Israel	Israel	MSQLI (including MSSS) EDSS	96.5% response rate ***	Mental dimension of health-related quality of life	Demographic and clinical variables	Correlation and linear hierarchical regression analysis	Correlation: A strong positive correlation was found between social support and MHQOL ($r = 0.58, p < 0.01$) Regression: Social support was a significant predictor of MHQOL beyond all of the other variables ($\beta = 0.4, p < 0.01$)
Suh et al. (2012)	218 participants with relapsing-remitting MS (mean duration 8 years, range 1-35; 197 females;	Recruited through an advertisement on the National Multiple Sclerosis Society website	USA	SPS HADS GLTEQ PDDS PSS	86.5% response rate ***	Depression	Physical activity, mobility disability and perceived stress	Bivariate correlation and multiple regression analysis	Correlation: A statistically significant correlation was found between social support and the HADS ($r = -0.386, p < 0.001$)

	mean age 43.5, range 17-70; 200 Caucasian)								Regression: Social support was a statistically significant predictor of depression ($\beta = -0.37, p = 0.0001$) for step one and $\beta = -0.14, p = 0.05$ for step two (when mobility disability and perceived stress entered the model)
Sullivan et al. (2004)	100 MS patients with a clinically definite or laboratory-supported definite diagnosis of MS (72 remission, 21 exacerbation, 7 secondary progressive; mean duration 8.54 years; 84 females; mean age 44.06; ethnicity: 80 White, 6	Recruited from the University of Maryland Medical Systems MS Center and the Neurology Center of Fairfax	USA	NSSQ COPE PAIS FIM	59.5% response rate ***	Psychosocial adjustment (healthcare orientation, vocational environment, domestic environment, sexual relationships, social environment and psychological distress)	Coping, uncertainty, demographic and clinical variables	Pearson product-moment correlation analysis and hierarchical regression analysis	Correlation: A statistically significant correlation was found between social support and adjustment ($r = -0.22, p < 0.05$) Regression: Social support was not found to be a statistically significant predictor of adjustment ($\beta = -0.12$)

	Black, 1 Other, 13 Missing)								
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*The data included in the Bamer et al. (2008) study includes a ‘western sample’ which is the same data set as the study conducted by Chwastiak et al. (2002) study. However, the Bamer et al. (2008) study also includes an ‘eastern sample’ of new data for comparison and therefore the studies have been reported separately.

**The data included in the Beal and Stuifbergen (2007) study is from the same pool of data as the Harrison and Stuifbergen (2002) study. However, the Beal and Stuifbergen study looked at a specific aspect of the CES-D (loneliness) and therefore the studies have been reported separately.

***The response rates for these studies were not directly provided in the paper but were calculable from the details provided. These response rates were calculated by determining the percentage of responses received from the number of requests sent (the number of ineligible participants was removed from both the number sent and returned, and the number of those who declined to participate were deducted from the amount received).

Note: **MSSS** = Medical Outcomes Study Modified Social Support Scale; **M-PS** = Mobility subscale of the Performance Scales; **PHQ-9** = Patient Health Questionnaire; **EDSS** = Expanded Disability Status Scale; **CES-D** = Center for Epidemiological Studies Depression Scale; **PRQ** = Personal Resource Questionnaire; **ISS** = Incapacity Status Scale; **MAI** = Multilevel Assessment Inventory; **DOI** = Demands of Illness Inventory; **SF36v2** = Medical Outcome Study 36-Item Health Survey Short Form (adapted to Portuguese); **SPS** = Social Provisions Scale; **RSES** = Rosenberg Self-Esteem Scale; **MSIS-29** = 29-item Multiple Sclerosis Impact Scale; **PDDS** = Patient Determined Disease Steps; **BDI** = Beck Depression Inventory; **MSQLI** = Multiple Sclerosis Quality of Life Inventory; **MusiQoL** – Multiple Sclerosis International Quality of Life Questionnaire; **BDI-II** = Beck Depression Inventory II; **BAI** = Beck Anxiety Inventory; **FSS** = Fatigue Severity Scale; **MHS** = Miller Hope Scale; **SSQ-6** = Social Support Questionnaire; **DSRS** = Depression Self-Rating Scale; **STAI** = State and Trait Anxiety Inventory; **SEI** – Self-Esteem Inventory; **TAS 20** = Toronto Alexithymia Scale; **CHIP** = Coping about Health Injuries and Problems; **MSRS** = MS-Related Scale; **MSRSS** = MS-Related Symptom Scale; **PSQ** = Postpartum Support Questionnaire; **PRQ 85** = Personal Resource Questionnaire; **CCS** = Concern for Children Scale; **CESD-10**; Center for Epidemiological Studies Depression 10-item scale); **MSPSS** – Multidimensional Scale of Perceived Social Support; **MSQOL-54** = Multiple Sclerosis Quality of Life Scale; **C-DRS** = Connor-Davidson Resilience Scale; **ED-A** = Emotional Distress-Anxiety Scale; **MCS** = Mental Component Summary; **SSF-WHOQOL-100** = Social Support Facet of World Health Organisation Quality of Life-100 Scale; **WOCQ** = Ways of Coping Questionnaire; **POMS-SF** = Profile of Mood States-Short Form; **PSSI – Family-based and Friend-based** = Perceived Social Support Inventory – Family-based and Friend-based; **WAIS** = Wechsler Adult Intelligence Scale (vocabulary subtest); **NSSQ** = Norbeck Social Support Questionnaire; **WCC** = Ways of Coping Checklist; **TSCS** = Tennessee Self-Concept Scale; **HADS** = Hospital Anxiety and Depression Scale; **GLTEQ** = Godin Leisure-Time Exercise Questionnaire; **PSS** = Perceived Stress Scale; **COPE** = COPE Inventory; **PAIS** = Psychosocial Adjustment to Illness Scale; **MUIS** = Mishel Uncertainty in Illness Scale; **FIM** = Functional Independence Measure.

Appendix 1-A: PRIMSA (2009) Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	

Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	

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Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	

DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit: www.prisma-statement.org.

Appendix 1-B: NICE Quality Appraisal Checklist (2012)

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Appendix G Quality appraisal checklist – quantitative studies reporting correlations and associations

A correlates review (see [section 3.3.4](#)) attempts to establish the factors that are associated or correlated with positive or negative health behaviours or outcomes. Evidence for correlate reviews will come both from specifically designed correlation studies and other study designs that also report on correlations.

This checklist^[15] has been developed for assessing the validity of studies reporting correlations. It is based on the appraisal step of the 'Graphical appraisal tool for epidemiological studies (GATE)', developed by Jackson et al. (2006).

This checklist enables a reviewer to appraise a study's internal and external validity after addressing the following key aspects of study design: characteristics of study participants; definition of independent variables; outcomes assessed and methods of analyses.

Like GATE, this checklist is intended to be used in an electronic (Excel) format that will facilitate both the sharing and storage of data, and through linkage with other documents, the compilation of research reports. Much of the guidance to support the completion of the critical appraisal form that is reproduced below also appears in 'pop-up' windows in the electronic version^[16].

There are 5 sections of the revised GATE. Section 1 seeks to assess the key population criteria for determining the study's **external validity** – that is, the extent to which the findings of a study are generalisable beyond the confines of the study to the study's source population.

Sections 2 to 4 assess the key criteria for determining the study's **internal validity** – that is, making sure that the study has been carried out carefully, and that the identified associations are valid and are not due to some other (often unidentified) factor.

Checklist items are worded so that 1 of 5 responses is possible:

++	Indicates that for that particular aspect of study design, the study has been designed or conducted in such a way as to minimise the risk of bias.
+	Indicates that either the answer to the checklist question is not clear from the way the study is reported, or that the study may not have addressed all potential sources of bias for that particular aspect of study design.

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–	Should be reserved for those aspects of the study design in which significant sources of bias may persist.
Not reported (NR)	Should be reserved for those aspects in which the study under review fails to report how they have (or might have) been considered.
Not applicable (NA)	Should be reserved for those study design aspects that are not applicable given the study design under review (for example, allocation concealment would not be applicable for case-control studies).

In addition, the reviewer is requested to complete in detail the comments section of the quality appraisal form so that the grade awarded for each study aspect is as transparent as possible.

Each study is then awarded an overall study quality grading for internal validity (IV) and a separate one for external validity (EV):

- ++ All or most of the checklist criteria have been fulfilled, where they have not been fulfilled the conclusions are very unlikely to alter.
- + Some of the checklist criteria have been fulfilled, where they have not been fulfilled, or not adequately described, the conclusions are unlikely to alter.
- – Few or no checklist criteria have been fulfilled and the conclusions are likely or very likely to alter.

Checklist

Study identification: Include full citation details	
Study design: <ul style="list-style-type: none"> • Refer to the glossary of study designs (appendix D) and the algorithm for classifying experimental and observational study designs (appendix E) to best describe the paper's underpinning study design 	
Guidance topic:	
Assessed by:	
Section 1: Population	

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1.1 Is the source population or source area well described? <ul style="list-style-type: none"> Was the country (e.g. developed or non-developed, type of health care system), setting (primary schools, community centres etc), location (urban, rural), population demographics etc adequately described? 	++ + – NR NA	Comments:
1.2 Is the eligible population or area representative of the source population or area? <ul style="list-style-type: none"> Was the recruitment of individuals, clusters or areas well defined (e.g. advertisement, birth register)? Was the eligible population representative of the source? Were important groups underrepresented? 	++ + – NR NA	Comments:
1.3 Do the selected participants or areas represent the eligible population or area? <ul style="list-style-type: none"> Was the method of selection of participants from the eligible population well described? What % of selected individuals or clusters agreed to participate? Were there any sources of bias? Were the inclusion or exclusion criteria explicit and appropriate? 	++ + – NR NA	Comments:
Section 2: Method of selection of exposure (or comparison) group		
2.1 Selection of exposure (and comparison) group. How was selection bias minimised? <ul style="list-style-type: none"> How was selection bias minimised? 	++ + – NR NA	Comments:
2.2 Was the selection of explanatory variables based on a sound theoretical basis? <ul style="list-style-type: none"> How sound was the theoretical basis for selecting the explanatory variables? 	++ + – NR NA	Comments:

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2.3 Was the contamination acceptably low? <ul style="list-style-type: none"> • Did any in the comparison group receive the exposure? • If so, was it sufficient to cause important bias? 	++ + – NR NA	Comments:
2.4 How well were likely confounding factors identified and controlled? <ul style="list-style-type: none"> • Were there likely to be other confounding factors not considered or appropriately adjusted for? • Was this sufficient to cause important bias? 	++ + – NR NA	Comments:
2.5 Is the setting applicable to the UK? <ul style="list-style-type: none"> • Did the setting differ significantly from the UK? 	++ + – NR NA	Comments:
Section 3: Outcomes		
3.1 Were the outcome measures and procedures reliable? <ul style="list-style-type: none"> • Were outcome measures subjective or objective (e.g. biochemically validated nicotine levels ++ vs self-reported smoking –)? • How reliable were outcome measures (e.g. inter- or intra-rater reliability scores)? • Was there any indication that measures had been validated (e.g. validated against a gold standard measure or assessed for content validity)? 	++ + – NR NA	Comments:
3.2 Were the outcome measurements complete? <ul style="list-style-type: none"> • Were all or most of the study participants who met the defined study outcome definitions likely to have been identified? 	++ + – NR NA	Comments:

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3.3 Were all the important outcomes assessed? <ul style="list-style-type: none"> • Were all the important benefits and harms assessed? • Was it possible to determine the overall balance of benefits and harms of the intervention versus comparison? 	++ + – NR NA	Comments:
3.4 Was there a similar follow-up time in exposure and comparison groups? <ul style="list-style-type: none"> • If groups are followed for different lengths of time, then more events are likely to occur in the group followed-up for longer distorting the comparison. • Analyses can be adjusted to allow for differences in length of follow-up (e.g. using person-years). 	++ + – NR NA	Comments:
3.5 Was follow-up time meaningful? <ul style="list-style-type: none"> • Was follow-up long enough to assess long-term benefits and harms? • Was it too long, e.g. participants lost to follow-up? 	++ + – NR NA	Comments:
Section 4: Analyses		
4.1 Was the study sufficiently powered to detect an intervention effect (if one exists)? <ul style="list-style-type: none"> • A power of 0.8 (i.e. it is likely to see an effect of a given size if one exists, 80% of the time) is the conventionally accepted standard. • Is a power calculation presented? If not, what is the expected effect size? Is the sample size adequate? 	++ + – NR NA	Comments:
4.2 Were multiple explanatory variables considered in the analyses? <ul style="list-style-type: none"> • Were there sufficient explanatory variables considered in the analysis? 	++ + – NR NA	Comments:

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4.3 Were the analytical methods appropriate? <ul style="list-style-type: none"> • Were important differences in follow-up time and likely confounders adjusted for? 	++ + – NR NA	Comments:
4.6 Was the precision of association given or calculable? Is association meaningful? <ul style="list-style-type: none"> • Were confidence intervals or p values for effect estimates given or possible to calculate? • Were CIs wide or were they sufficiently precise to aid decision-making? If precision is lacking, is this because the study is under-powered? 	++ + – NR NA	Comments:
Section 5: Summary		
5.1 Are the study results internally valid (i.e. unbiased)? <ul style="list-style-type: none"> • How well did the study minimise sources of bias (i.e. adjusting for potential confounders)? • Were there significant flaws in the study design? 	++ + –	Comments:
5.2 Are the findings generalisable to the source population (i.e. externally valid)? <ul style="list-style-type: none"> • Are there sufficient details given about the study to determine if the findings are generalisable to the source population? • Consider: participants, interventions and comparisons, outcomes, resource and policy implications. 	++ + –	Comments:

^[15] Appraisal form derived from: Jackson R, Ameratunga S, Broad J et al. (2006) The GATE frame: critical appraisal with pictures. Evidence Based Medicine 11: 35–8.

^[16] Available from CPHE on request.

Appendix 1-C: Quality Assessment Scoring Description

- A score of ++ suggests that the aspect of study design has been conducted to minimise the risk of bias.
- A score of + suggests that the answer to the checklist was either not clear from the reporting of the study, or that all potential sources of bias for that aspect of design may not have been addressed.
- A score of - showed that the aspect of study design contained significant sources of bias which may endure.
- A score of NR (not reported) suggests that the aspect of design being reviewed failed to report how they have (or might have) been considered.
- A score of NA (not applicable) suggests that the aspect of design being reviewed was not applicable given the study design.

For the final two summary questions there are three possible responses:

- A score of ++ suggests that all or most of the checklist criteria were fulfilled, or conclusions are very unlikely to alter.
- A score of + suggests that some of the checklist criteria have been fulfilled or that the conclusions are unlikely to alter.
- A score of - suggests that few or no checklist criteria were fulfilled and conclusions are likely or very likely to alter.

Appendix 1-D: Excluded Articles

Study	Reason for Exclusion
Anagnostouli, M., Katsavos, S., Artemiadis, A., Zacharis, M., Argirou, P., Theotoka, I., ... & Stamboulis, E. (2014). Determinants of stigma experienced by patients suffering from multiple sclerosis. A Hellenic population study. <i>Multiple Sclerosis</i> , 20(1: supplement 1), 69.	Not assessing relationship between social support and psychological variable
Anens, E., Zetterberg, L., Urell, C., Emtner, M., & Hellström, K. (2017). Self-reported physical activity correlates in Swedish adults with multiple sclerosis: a cross-sectional study. <i>BMC Neurology</i> , 17(1), 204.	Not assessing relationship between social support and psychological variable
Arnett, P. A., Barwick, F. H., & Beeney, J. E. (2008). Depression in multiple sclerosis: review and theoretical proposal. <i>Journal of the International Neuropsychological Society</i> , 14(5), 691-724.	Review paper
Briones-Buixassa, L., Milà, R., Aragonès, J. M., Bufill, E., & Arrufat, F. X. (2015). The role of psychosocial moderator factors in the relationship between stress and multiple sclerosis: a case-control study. <i>Psychotherapy & Psychosomatics</i> , 84(supplement 1), 10.	Conference paper
Cadden, M. H., Arnett, P. A., Tyry, T. M., & Cook, J. E. (2018). Judgment hurts: The psychological consequences of experiencing stigma in multiple sclerosis. <i>Social Science & Medicine</i> , 208, 158-164.	Not assessing relationship between social support and psychological variable
Gatten, C. W., Brookings, J. B., & Bolton, B. (1993). Mood fluctuations in female multiple sclerosis patients. <i>Social Behavior & Personality: an International Journal</i> , 21(2), 103-106.	Wrong construct of social support
Geertz, W., Dechow, A. S., Patra, S., Heesen, C., Gold, S. M., & Schulz, K. H. (2015). Changes of motivational variables in patients with multiple sclerosis in an exercise intervention: associations between physical performance and motivational determinants. <i>Behavioural Neurology</i> , 2015(248193), 7 pages.	Wrong construct of social support
Gulick, E. E. (2001). Emotional distress and activities of daily living functioning in persons with multiple sclerosis. <i>Nursing Research</i> , 50(3), 147-154.	Wrong construct of social support
Hoover, M. H. (2005). The relationship between stress and symptomatology of Multiple Sclerosis moderated by perceived marital support. <i>ProQuest Information & Learning</i> , 66, 1776	Wrong construct of social support
Hwang, J. E., Cvitanovich, D. C., Doroski, E. K., & Vajarakitipongse, J. G. (2011). Correlations between quality of life and adaptation factors among people with multiple sclerosis. <i>American Journal of Occupational Therapy</i> , 65(6), 661-669.	Wrong construct of social support
James, E., Mills, R. J., Tennant, A., & Young, C. A. (2016). Identification of coping strategies in Multiple Sclerosis. <i>Multiple Sclerosis</i> , 22(supplement 3), 432	Conference paper
Kisvetrova, H., Kralova, J. (2014). Basic factors influencing death anxiety. <i>Ceskoslovenska Psychologie</i> , 58(1), 41-51	Not in English
Kleiboer, A. M., Kuijer, R. G., Hox, J. J., Jongen, P. J., Frequin, S. T., & Bensing, J. M. (2007). Daily negative interactions and mood among patients and partners dealing with multiple sclerosis (MS): The	Wrong construct of social support

moderating effects of emotional support. <i>Social Science & Medicine</i> , 64(2), 389-400.	
Korostil, M., & Feinstein, A. (2007). Anxiety disorders and their clinical correlates in multiple sclerosis patients. <i>Multiple Sclerosis Journal</i> , 13(1), 67-72.	Wrong construct of social support
Long, D. D., & Miller, B. J. (1991). Suicidal tendency and multiple sclerosis. <i>Health & Social Work</i> , 16(2), 104-109.	Wrong construct of social support
Maybury, C. P., & Brewin, C. R. (1984). Social relationships, knowledge and adjustment to multiple sclerosis. <i>Journal of Neurology, Neurosurgery & Psychiatry</i> , 47(4), 372-376.	Not assessing relationship between social support and psychological variable
Mohr, D. C., Classen, C., & Barrera, M. (2004). The relationship between social support, depression and treatment for depression in people with multiple sclerosis. <i>Psychological Medicine</i> , 34(3), 533-541.	Not assessing relationship between social support and psychological variable
Montanari, E., Pattini, M., Cenci, F., Pesci, I., Manneschi, L., Guareschi, A. ... Marta, E. (2009). Quality of life in multiple sclerosis: Social support and depression. <i>Multiple Sclerosis</i> , 15(9: Supplement S), S266	Presentation abstract
Paddam, A., Barnes, D., Langdon, D. (2009). Why are multiple sclerosis patients angry? A preliminary quantitative study of factors associated with anger in multiple sclerosis. <i>Multiple Sclerosis</i> , 15(9: Supplement S), S228	Presentation abstract
Pattini, M., Cenci, F., Allegri, B., Pesci, I., Manneschi, L., Guareschi, A. ... Montanari, E. (2010). Quality of life in multiple sclerosis: Social support, depression, personality and coping style. <i>Multiple Sclerosis</i> , 16(10: Supplement 1), S180-S181	Presentation abstract
Phillips, L. J., & Stuifbergen, A. K. (2010). The relevance of depressive symptoms and social support to disability in women with multiple sclerosis or fibromyalgia. <i>International Journal of Rehabilitation Research</i> , 33(2), 142.	Not assessing relationship between social support and psychological variable
Rommer, P. S., Sühnel, A., König, N., & Zettl, U. K. (2017). Coping with multiple sclerosis—the role of social support. <i>Acta Neurologica Scandinavica</i> , 136(1), 11-16.	Wrong construct of social support
Siegel, S. D., Turner, A. P., & Haselkorn, J. K. (2008). Adherence to disease-modifying therapies in multiple sclerosis: does caregiver social support matter?. <i>Rehabilitation Psychology</i> , 53(1), 73.	Wrong construct of social support
Strober, L. B. (2016). A biopsychosocial approach to understanding quality of life in early onset multiple sclerosis (MS). <i>Quality of Life Research</i> , 25(1: Supplement 1), 152.	Conference paper
Strober, L. B. (2018). Quality of life and psychological well-being in the early stages of multiple sclerosis (MS): Importance of adopting a biopsychosocial model. <i>Disability and Health Journal</i> , 11(4), 555-561.	Not assessing relationship between social support and psychological variable
Terzi, M., Sen, S., Kumcagiz, H., Terzi, Y. (2018). Variables that affect life satisfaction of multiple sclerosis patients a comparative study. <i>Multiple Sclerosis Journal</i> , 24(3), 387.	Conference paper
Warren, S., Warren, K. G., & Cockerill, R. (1991). Emotional stress and coping in multiple sclerosis (MS) exacerbations. <i>Journal of</i>	Not assessing relationship between

<i>Psychosomatic Research</i> , 35(1), 37-47.	social support and psychological variable
Wilski, M., Tomczak, M. (2016). Illness perception in multiple sclerosis. Sociodemographic, illness-related, and psychosocial correlates. <i>Multiple Sclerosis</i> , 22(Supplement 3), 822.	Presentation abstract
Wineman, N. M. (1990). Adaptation to multiple sclerosis: the role of social support, functional disability, and perceived uncertainty. <i>Nursing research</i> , 39(5), 294-299	Wrong construct of social support
Wollin, J. A., Fulcher, G., McDonald, E., Spencer, N., Mortlock, M. Y., Bourne, M., & Simmons, R. D. (2010). Psychosocial factors that influence quality of life and potential for self-management in multiple sclerosis. <i>International Journal of MS Care</i> , 12(3), 133-141.	Wrong construct of social support
Wollin, J. A., Spencer, N., McDonald, E., Fulcher, G., Bourne, M., & Simmons, R. D. (2013). Longitudinal changes in quality of life and related psychosocial variables in Australians with multiple sclerosis. <i>International Journal of MS Care</i> , 15(2), 90-97.	Wrong construct of social support
Yamout, B., Issa, Z., Herlopian, A., El Bejjani, M., Khalifa, A., Ghadieh, A. S., & Habib, R. H. (2013). Predictors of quality of life among multiple sclerosis patients: a comprehensive analysis. <i>European Journal of Neurology</i> , 20(5), 756-764.	Wrong construct of social support

Appendix 1-E: Duplicate Samples

Four studies used the same data sets within the studies; the data included in the Bamer et al. (2008) study includes a ‘western sample’ ($n = 661$) which is from the same data set as the study conducted by Chwastiak et al. (2002) study. However, the Bamer et al. (2008) study also includes an ‘eastern sample’ of new data for comparison and therefore the studies have been reported separately. Moreover, the data included in the Beal and Stuifbergen (2007) study ($n = 659$) is from the same pool of data as the Harrison and Stuifbergen (2002) study. However, the Beal and Stuifbergen (2007) study looked at a specific aspect of the CES-D (loneliness) and therefore the studies have been reported separately.

Appendix 1-F: Author Guidelines



BJHP AUTHOR GUIDELINES

Sections

1. [Submission](#)
2. [Aims and Scope](#)
3. [Manuscript Categories and Requirements](#)
4. [Preparing the Submission](#)
5. [Editorial Policies and Ethical Considerations](#)
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9. [Editorial Office Contact Details](#)

1. SUBMISSION

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Author Guidelines updated April 2019

Section 2: Empirical Paper

Does a lack of social support and perceived stigma contribute to psychological distress in individuals with motor neurone disease?

Natalie Leigh*

Doctorate in Clinical Psychology, Lancaster University

Word count (exc. title page, abstract, appendices, figures and tables): 7,584

*Requests for reprints should be addressed to Natalie Leigh, Doctorate in Clinical Psychology, Division of Health Research, Furness Building, Lancaster University, Lancaster, LA1 4YG, United Kingdom (e-mail: n.leigh@lancaster.ac.uk)

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Abstract

Objectives: This study aimed to identify whether a lack of social support and increased levels of perceived stigma predicted psychological distress for individuals with a diagnosis of motor neurone disease (MND) also known as amyotrophic lateral sclerosis. Although identified in other neurodegenerative conditions, and in relation to quality of life for MND, social support and stigma have not previously been investigated as predictors of psychological distress in people with MND. **Design:** A cross-sectional design utilising an online survey method was used. It was hypothesised that both social support and stigma would be significant predictors of psychological distress, over and above demographic and clinical variables. **Methods:** Individuals with a diagnosis of MND were recruited internationally through social media and through various organisations and support services for people with MND. Seventy-seven participants completed the survey and data were analysed using hierarchical regression analyses. **Results:** Significant correlations were identified between social support, felt and enacted stigma and psychological distress. Regression analyses revealed that enacted stigma was not an independent predictor in any of the models and social support did not remain a significant independent predictor for stress when stigma entered the model. However, felt stigma was a significant independent predictor in all the models and was a more powerful predictor than social support in each of the models. **Conclusions:** Stigma and social support may be important to consider for ameliorating psychological distress for people with MND. Limitations of the current study are discussed, along with implications for clinical practice.

Keywords: Social support; stigma; psychological distress; motor neurone disease; amyotrophic lateral sclerosis

Introduction

Motor neurone disease (MND), also referred to as amyotrophic lateral sclerosis (ALS) and Lou Gehrig disease in the US, is a life-limiting neurodegenerative condition. The condition progressively destroys the motor neurons in the brain and spinal cord and alters an individual's ability to control voluntarily their muscle movements, leading to paralysis, swallowing difficulties, respiratory failure and ultimately, death (King, Mulligan & Stansfield, 2014). The effects of MND are not limited to motor functions; behaviour difficulties, cognitive impairment (McCluskey et al., 2009; Strong et al., 1999) emotional difficulties (depression, anxiety and anger) and involuntary changes in mood (Orrell, 2016) are also common.

Characteristically, MND is more common in men than women (Stone, 1987) and the average age of onset is between 60 and 65 years of age, with the likelihood of developing the condition increasing with age (Talbot & Marsden, 2008). The prevalence rate for North America and Europe is around two per 100,000 of the population (Worms, 2001) and median survival rates following symptom onset are generally only two to four years, with only 10%-20% of individuals surviving past 10 years (Chiò et al., 2009).

Current interventions for individuals with MND mainly focus upon the physical aspects of the condition, to maintain physical and biological functioning and quality of life for as long as possible (Andersen et al., 2012). However, a review (McLeod & Clarke, 2007) highlighted the lack of guidance on psychological care and the review authors proposed that, given a range of psychosocial issues impact on the quality of life of individuals with MND, a more holistic approach to care was required. A quantitative and qualitative review, 10 years later, assessed literature aimed at the supportive care needs of individuals with MND and their caregivers. Of the 37 studies included, 16 discussed psychological needs, 18 discussed

social needs and 13 discussed emotional needs. Despite more research recognising these needs of individuals with MND and their caregivers, the review highlighted that there is still a significant need for more psychological, social and emotional support, alongside physical and practical support (Oh & Kim, 2017). They proposed that the psychological impact of receiving and adjusting to this diagnosis along with the impact of coping with the associated functional changes should be considered equally, in line with the physical impact of the condition to enhance and maintain an individual's quality of life. Higher levels of psychological well-being may also be protective for physical health as quantitative research suggests that individuals with a higher level of well-being may have a lower risk of mortality, even when disease severity and length of illness are controlled (McDonald, Wiedenfeld, Hillel, Carpenter & Walter, 1994). However, even when psychological needs of low mood, anxiety and involuntary changes in mood are identified, management strategy literature, such as that produced by Gordon (2011), mainly refers to the use of medication as treatment options, although the use of psychotherapy was mentioned. This indicates that psychological and social understandings and interventions are not always considered as the main treatment options when considering the psychological needs of this population.

Due to the challenges that the condition presents, individuals with a diagnosis of MND are likely to experience psychological distress and decreased well-being (Hogg, Goldstein & Leigh, 1994; Lou, Reeves, Benice & Sexton, 2003; Montgomery & Erikson, 1987; Tedman, Young & Williams, 1997; Vignola et al., 2008). Although several factors undoubtedly contribute to this (e.g., biological, social, psychological and spiritual suffering: Ganzini, Johnston & Hoffman, 1999; social withdrawal: Rigby et al., 1999 and physical impairment: Hunter, Robinson & Neilson, 1993; Hogg et al., 1994), recent research in other physical health conditions has suggested that stigma may be one important variable to consider. This has been identified as important in health conditions with perceivable physical

effects such as, epilepsy (Baker, Eccles & Caswell, 2018), Parkinson's disease (Maffoni, Giardini, Pierobon, Ferrazzoli & Frazzitta, 2017) and multiple sclerosis (Cadden, Arnett, Tyry & Cook, 2018; Broersma, Oeseburg, Dijkstra & Wynia, 2018).

The term stigma was originally defined by Goffman (1963) as “an undesired differentness” (p. 5) and this has since been developed to include two concepts of stigma; felt and enacted (Scambler & Hopkins, 1986). ‘Felt stigma’ refers to a feeling of shame about being different and feeling that discrimination for this difference will occur, whereas ‘enacted stigma’ refers to actual experience of this discrimination. This definition has since been developed and Link and Phelan (2001) described stigma as a set of components which include labelling, stereotyping, separation, status loss, and discrimination. Therefore, the term stigma can encompass a range of negative actions and associations that can be attributed to an individual based on their differentness.

The influence of stigma as a predictor of quality of life in individuals diagnosed with neuromuscular conditions including MND, has been quantitatively studied by van der Beek, Bos, Middel and Wynia (2013). This study found that stigma was a major predictor of poorer quality of life, with ‘felt stigma’ being a stronger predictor compared with ‘enacted stigma’. However, this study focused on quality of life, rather than psychological distress in particular, and individuals diagnosed with MND only made up 9% of the total number of participants. Due to the life-limiting nature of MND in comparison to the other conditions included within the study (muscle disorder, junction disorder and peripheral nerve disorder), it seems important to study the effects of stigma in this population independently.

While extensively researched in other neurodegenerative conditions such as multiple sclerosis (Bambara, Turner, Williams & Haselkorn, 2011; Bamer, Cetin, Johnson, Gibbons & Ehde, 2008; Chwastiak et al., 2002; Harrison & Stuifbergen, 2002; Suh, Weikert, Dlugonski,

Sandroff & Motl, 2012) and Parkinson's disease (Ghorbani Saeedian et al., 2014; Cheng et al., 2008), the impact of social support as a predictor of psychological distress for individuals with MND has only been assessed with a very small sample. Matuz, Birbaumer, Hautzinger and Kübler (2010) identified perceived social support as a significant predictor of depression and quality of life for individuals with a diagnosis of MND which included a sample of 27 participants. Matuz, Birbaumer, Hautzinger and Kübler (2015) conducted a longitudinal study and found that social support was a significant predictor of depression and quality of life at a subsequent time point for this population. However, only 27 participants were included at the initial time point, which reduced to 16 for the final time point. Social support has also been identified as a correlate of quality of life for individuals with a diagnosis of MND (Ganzini, et al., 1999; Goldstein, Atkins & Leigh, 2002; Simmons, Bremer, Robbins, Walsh & Fischer, 2000). However, these studies used restricted measures of social support which only assessed the impact of close relationships, rather than broader social support beyond the household (i.e. other family members or friends) and did not include a regression model to identify the predictive nature of social support, controlling for other variables.

Evidence does suggest, however, that social support is an issue of concern for people with MND. Mistry and Simpson (2013) conducted a qualitative study exploring the process of receiving a diagnosis of MND and discovered that functional changes caused by MND can impact on an individual's social engagement, social status and identity, affecting an individual's relationships with family and friends. Moreover, it has also been qualitatively noted by Cobb and Hamera (1986) that social relationships undergo radical changes following a diagnosis of MND. It therefore feels important also to quantitatively assess the relationship of stigma and social support on levels of psychological distress in individuals with MND. Moreover, social support has already been identified as a correlate of quality of life and as a significant predictor of depression in this population and stigma has been

identified as an important predictor of psychological distress in different physical health conditions, including neurodegenerative conditions. Therefore, determining the role that each of these variables has on levels of psychological distress in people with MND, will build on past research and address a current gap in this area of literature.

Consequently, this study aims to investigate the relationship between the lack of social support and perceived stigma on psychological distress for people with a diagnosis of MND. The findings could highlight whether it is important to consider these factors when assessing psychological distress in practice and to inform interventions. It also aims to discover the strength of these relationships when controlling for demographics and levels of physical functioning. The specific research question that this study aims to answer is: Do lower levels of social support and increased levels of perceived stigma contribute to psychological distress in individuals with MND? It is hypothesised that stigma and social support will each have a significant effect over and above the demographic and physical functioning variables in predicting psychological distress (depression, anxiety and stress) for individuals with a diagnosis of MND.

Method

Design

A quantitative cross-sectional survey design was used to investigate whether perceived stigma and social support were significant predictors of psychological distress (depression, anxiety and stress) in individuals with a diagnosis of MND. The specific hypothesis for this study was that both perceived stigma and social support would be significant predictors of psychological distress in individuals with a diagnosis of MND, over and above demographic and clinical variables.

The regression models were theoretically driven and based on previous research. Thus, the variables included in the hierarchical block regression analysis were entered as follows: demographics (age and gender); clinical variable (physical functioning); social support and stigma (felt and enacted). Social support and stigma were entered separately to identify their individual relationships. Social support was entered before stigma as this variable had already been identified as a predictor of depression and quality of life in individuals with MND, whereas stigma was a new variable to be considered as this had not been studied before exclusively with people with MND.

Participants

Individuals aged 18 years or over who had a diagnosis of MND/ ALS/ Lou Gehrig disease and who could complete an online survey written in English (either alone or with assistance from another person) were eligible to take part. An opportunistic sampling method was employed as participants volunteered to participate following advertisement of the study details online, using social media and through international organisations. If a participant wished to take part in the study, then they accessed the study link provided in the study advert and were given the opportunity to read and download the participant information sheet. Eligibility for the study was based on self-report and was recorded through a demographic questionnaire at the beginning of the survey.

An *a priori* power calculation (using G*Power software based on 5-8 predictors, presuming a medium effect size of 0.15) suggested between 92 and 109 participants were required (80% power, $\alpha = .05$). A total of 77 participants were recruited; 34 females and 43 males. The recruitment method employed received 134 responses from participants, but of these, only 94 opened the survey and proceeded further. Of those who proceeded further, 84 completed demographic data, with only 80 of these continuing to complete the outcome measures. Of the 80 who completed the outcome measures, only 78 completed all the

measures (two participants did not complete the physical functioning measure and as such their data were withdrawn). A further participant's data were withdrawn due to not meeting the inclusion criteria of being completed by an individual with a diagnosis of MND, leaving a total of 77 participants whose data were included in the final analysis.

Please see Table 1 for participants' self-reported demographic characteristics.

Insert Table 1 here please

Procedure

Ethical approval was gained from the principal investigator's host academic institution (please see section 4 of the thesis for full details).

All the study documents were reviewed by a family member of an individual who had a diagnosis of MND and by a service-user from the academic institution's public involvement network who had an interest in this field. The feedback gained from this consultation was used to decide on the measures and to review the wording of the documentation to ensure it was appropriate for the audience. They also provided feedback on the duration of the study to ensure this would not be too burdensome for the participants. This consultation preceded finalisation of the documents and submission for ethical approval as part of the research development process.

Recruitment for the study took place online from a variety of sources, mainly through international organisations supporting people with MND. This was achieved by sharing the study advert and link on social media (Facebook and Twitter) pages by the Motor Neurone

Disease Association (MNDA) Australia, MNDA UK, MNDA New Zealand, MNDA Scotland, ALS Association Canada, ALS Society Quebec, Minds and Movement, Lancaster Centre for Ageing Research and the principal investigator. Furthermore, the information was shared through newsletters, (MNDA UK and MNDA South Africa), through the research page of their website (MNDA UK), through staff contact with individuals with a diagnosis of MND (MNDA Scotland and the Les Turner Foundation – based in the USA) and through research databases (New Zealand MND Registry and MNDA South Africa). The New Zealand MND Registry was established in 2017 to aid in the recruitment of participants for research projects and in July 2018 they had 142 participants enrolled on their database expressing an interest in participating in research (Walker et al., 2019), therefore they were able to provide a useful contribution to the recruitment process.

An online survey was constructed using Qualtrics software which was used to collect the data. Prior to the commencement of the survey each participant was shown the participant information sheet and had the chance to download this for their own reference. Participants had to complete the consent form prior to being allowed to start the survey. Following completion of each of the questionnaires, a debrief sheet was shown to each participant and they had the opportunity to download this for their own reference. Both the participant information sheet (see appendix 4-B of ethics section) and debrief sheet (see appendix 4-D of ethics section) informed participants that the information provided was anonymous, meaning that no personal identifiable information was held and therefore it was not possible to withdraw data once the survey had begun as an individual's data could not be identified. The personal details of any participants who requested to access a copy of the results were held long enough to send out this information, then were destroyed and this information was kept confidential throughout the process. Both the participant information

sheet and debrief sheet provided the contact details of organisations who could provide support if participation in the study caused any distress for individuals.

Measures

To control for potential confounders, measures were included to assess demographic variables (age and gender) and a clinical variable (physical functioning), along with the two variables of interest (social support and stigma). One measure was used to assess the outcome of psychological distress (measuring stress, anxiety and depression).

To situate the sample additional variables were collected including nationality, relationship status, time since symptom onset and time since diagnosis.

Demographic and Clinical data

The survey included a self-report questionnaire requesting the participants' demographic characteristics (age, gender, nationality and relationship status) and some clinical characteristics (time since symptom onset and time since diagnosis). The following were also administered:

Physical Functioning

The Self-Administered Amyotrophic Lateral Sclerosis Functional Rating Scale Revised (SA-ALSFRS-R; Cedarbaum et al., 1999; Montes et al., 2006) includes 12 questions which assess the domains of motor function, bulbar symptoms and breathing ability in individuals with MND. A scoring sheet is provided to score individual item responses on a scale of 0 to 4 with a total score range of 0 to 48, with higher scores indicating higher levels of physical functioning. Although there is no current evidence regarding the reliability and validity of the SA-ALSFRS-R, there is evidence regarding the clinician administered ALSFRS-R. This has been shown to be a reliable and valid measure by the authors during

development (Cedarbaum et al., 1999). Internal reliability was displayed by Cronbach's coefficient alpha score of 0.73 for the total scale. Montes et al. (2006) compared the use of the SA-ALSFRS-R to the clinician administered ALSFRS-R and reported an intraclass correlation coefficient score of $r = 0.93$, implying that the self-administered version is as reliable as the clinician administered version. This measure was chosen as it is aimed specifically at individuals with a diagnosis of MND to assess their physical functioning and symptom severity.

The use of the ALSFRS-R instrument online compared to on-site face-to-face assessment was assessed by Maier et al. (2012) and their results supported the use of the measure online, due to a highly significant correlation between on-site evaluation and online testing ($r = 0.96$). The SA-ALSFRS-R was developed specifically for use with individuals with a diagnosis of MND and is therefore suitable for use with this population.

Social Support

The Multidimensional Scale of Perceived Social Support (MSPSS; Zimet, Dahlem, Zimet & Farley, 1988) is a 12-item measure scored on a scale from 1 (very strongly disagree) to 7 (very strongly agree) with a total score range of 12 to 84 with four questions for each of the three subscales: significant other, family and friends (scores range from 4 to 28 for each subscale). Scores can be used for the total scale and for the subscales, with higher scores indicating higher levels of social support. This is a valid and reliable measure as reported by the authors. Cronbach's coefficient alpha scores are given as 0.91 for the significant other subscale, 0.87 for the family subscale, 0.85 for the friends subscale and 0.88 for the total scale. Test-retest reliability for each of the significant other, family, and friends subscales was 0.72, 0.85, and 0.75 respectively and the total scale was 0.85. This was chosen to assess social support because this is a relatively short and easy to complete measure, reducing

participant burden. It is a measure which provides an insight into an individual's levels of social support from different sources, providing subscales for these in relation to family, friends and significant others.

To the best of the authors knowledge, this measure has not previously been used online, nor with people with MND, however, it has been used with individuals with multiple sclerosis (Hyarat, Al-Gamal & Rama, 2018; Koelmel, Hughes, Alschuler & Ehde, 2017).

Stigma

The Stigma Scale for Chronic Illness (SSCI, Rao et al., 2009) comprises 24 questions with two subscales; one scale for felt stigma (labelled self-stigma) and one scale for enacted stigma. There are 13 questions for the felt stigma scale which relate to an individual's fear of discrimination and 11 questions for the enacted stigma scale which includes questions relating to their actual experience of discrimination in relation to illness. Each item is scored on a scale of 0 (never) to 4 (always) with a total score range of 0 – 96 for the full scale, between 0 – 52 for the felt scale and between 11 – 44 for the enacted scale. Higher scores indicate a higher level of stigmatisation. Internal reliability for this scale was assessed by the authors during development, and it was found to be a reliable measure with promising psychometric properties (Cronbach's coefficient alpha was 0.97 for the full-scale, with both subscales correlating strongly $r = 0.81$). This measure has been used in this study to assess levels of perceived stigma and was chosen because it has been specifically developed for people with chronic neurological illnesses, including individuals with MND. The total score was used alongside the two subscales (felt and enacted stigma) for the correlational analyses for this study and the two subscales were used in the regression analyses of this study.

The authors of this measure used it online during the validation on the psychometric properties of the measure (Rao et al., 2009) indicating its feasibility for online use. During

validation of the measure, participants with a diagnosis of MND were included, providing evidence for its use with this population.

Psychological Distress

The Depression, Anxiety and Stress Scale (DASS-21; Lovibond & Lovibond, 1995) has 21 questions scored across three subscales, depression, anxiety and stress, with seven questions for each subscale. These are scored on a range of 0 (never) to 3 (almost always) with a total range of 0 – 21 for each of the subscales. Higher scores indicate a higher level of distress and scores can be categorised as: “normal”, “mild”, “moderate”, “severe” or “extremely severe” ranges. This has been found to be an internally reliable measure, with Cronbach’s coefficient alphas of 0.94 and 0.88 reported for the depression subscale, 0.87 and 0.82 for the anxiety subscale and 0.91 and 0.90 for the stress subscale (Antony, Bieling, Cox, Enns, Swinson and Haynes, 1998; Henry and Crawford, 2005). This was selected to assess levels of psychological distress because it captures three forms of psychological distress within one measure, but with three subscales, it is non-combinable and relatively short to complete.

This measure has been used online in previous research with a sample of community-based carers (Farrugia, Hewitt, Bourke-Taylor & Joosten, 2019). This measure has been used to measure psychological distress in other research in individuals with MND (Lillo, Mioshi, Zoing, Kiernan & Hodges, 2011; Caga, Ramsey, Hogden, Mioshi & Kiernan, 2015) as well as individuals with Parkinson’s disease (Troeng, Egan & Gasson, 2014; Whitworth et al., 2013).

Data Analysis

Data analysis was completed using SPSS Version 25. The sample was checked for missing data prior to any analysis being conducted, and seven participants had not provided a

response to the same question on the SSCI: “people with my illness lost their jobs when their employers found out”. This missing datum was appropriately replaced with a mean value of 2 based on the mean of the specific subscale of the SSCI (enacted subscale). No other missing data was identified in the sample. Outliers were identified using boxplots and scores were checked for errors. The only outliers identified were for variables which were not included in the final analyses therefore no further action was required. All data were first checked for normality by using the Kolmogorov-Smirnov test, which yielded significant results, so a less conservative method of dividing the skewness or kurtosis value by its standard error was used due to the small sample size (Field, 2005). Using parameters of -3 to +3 the only scale not normally distributed for skewness and kurtosis was the MSPSS total score.

As the MSPSS total score was not normally distributed, non-parametric correlations were calculated using Spearman’s correlation coefficients to identify relationships between the variables. Then a theoretically driven hierarchical block regression was conducted and entry into regression was based on theoretical grounds. The predictors were entered in a stepwise manner, in four blocks, based on their importance as identified from previous research: 1) demographics (age and gender), 2) clinical variable (physical functioning) 3) social support and 4) stigma (felt and enacted). Three regression models were conducted – one for each of the three outcome variables: stress, anxiety and depression.

The Durbin-Watson statistic was checked for each of the models to determine whether the assumption of the independent errors was tenable, and all these values fell within the limits of +1 to +3. Variance inflation factors (VIF) were also checked and all of these were below 10, with the average VIF values being near the one mark (range = 1.009 – 1.945). Different rules of thumb have been advocated for VIFs (e.g., <10 or <4: see O’Brien, 2007) but with the average VIF value for these analyses being 1.195, multicollinearity was not a

concern. The assumptions of the model were also checked by assessing the scatterplot of the standardised residuals against the standardised predicted values, the histogram of standardised residuals and the normal probability plots. These showed that the assumptions of linearity and homoscedasticity were met and that the standardised residuals were normally distributed. All standardised residuals fell within -3.29 to +3.29 suggesting there were no outliers and the model fitted the data (Field, 2013).

Results

Participants

The demographic and clinical characteristics of these participants can be viewed in Table 1. More participants were male ($n = 43$) and the mean age of the sample was 59 years. The most common nationality reported by participants was 'New Zealander' ($n = 18$) and most participants were married ($n = 55$). Time since symptom onset had a median duration of 3.5 years (mean 4.75 years) and time since diagnosis had a median duration of 2.4 years (mean 3.53 years). Most participants did not require carer assistance to complete the survey ($n = 70$).

The mean scores, standard deviations and Cronbach's alpha scores for each measure are reported in Table 2.

Insert Table 2 here please

The mean (*SD*) score for the SA-ALSFRS-R was 29 (8.62), indicating that levels of independent functioning were within the mid-point range (minimum score is 0 and maximum score is 48, with higher scores indicating higher levels of independent functioning). The

majority of scores fell within the ‘mild to moderate’ category (43%, $n = 33$), the next common category was ‘moderate to severe’ (30%, $n = 23$), followed by the ‘advanced disease’ category (16%, $n = 12$) and finally the ‘minimal to mild’ category (12%, $n = 9$).

The mean (SD) score for the MSPSS total was 66.47 (15.66), indicating that levels of social support were towards the higher end of the scale for this sample (minimum score is 12 and maximum score is 84, with higher scores indicating higher levels of social support). The mean (SD) score for the SSCI total score was 32.83 (16.12), indicating that levels of stigma were quite low in this sample (minimum score is 0 and maximum score is 96, with higher scores indicating higher levels of stigma). For the SSCI self-subscale (measuring self-stigma) the mean (SD) score was 22.48 (10.36), and for the SSCI enacted-subscale the mean (SD) score was 10.09 (7.09).

The mean (SD) scores for the outcome variables (stress, anxiety and depression) were 7.03 (4.59), 5.32 (3.85) and 7.19 (5.21) respectively. For the stress subscale, the majority of scores were in the ‘normal’ category (62%, $n = 48$), the next most common category was ‘moderate’ (22%, $n = 17$), followed by the ‘mild’ category (9%, $n = 7$), followed by the ‘severe’ category (5%, $n = 4$) and finally the ‘extremely severe’ category (1% $n = 1$). For the anxiety subscale, all the scores fell within the ‘normal’ category (100%, $n = 77$). For the depression subscale, the majority of scores were in the ‘normal’ category (70%, $n = 54$), the next most common category was ‘mild’ (18%, $n = 14$), followed by the ‘moderate’ category (12%, $n = 9$). This indicates that the majority of the sample had ‘normal’ levels of stress, anxiety and depression.

Correlations

Spearman’s rho correlation coefficients were calculated to determine the relationships between the variables; see Table 3 for the results of these correlations.

Insert Table 3 here please

None of the demographic variables or the clinical variable correlated significantly with the outcome variables of stress, anxiety and depression. Statistically significant relationships were found between the main predictors of stigma and social support and all three outcome variables.

Social support (total score) was negatively correlated with all the DASS-21 scales (stress: $r_s = -0.385$, $p = 0.01$; anxiety: $r_s = -0.399$, $p = 0.01$; depression: $r_s = -0.437$, $p = 0.01$) indicating that less social support is associated with higher levels of stress, anxiety and depression. The social support total score was also negatively correlated with the stigma total score ($r_s = -0.483$, $p = 0.001$).

Stigma (total score) was positively correlated with all the DASS-21 scales (stress: $r_s = 0.538$, $p = 0.01$; anxiety: $r_s = 0.447$, $p = 0.01$; depression: $r_s = 0.660$, $p = 0.01$). The stigma self-subscale (measuring felt stigma) was also positively correlated with all the DASS-21 scales (stress: $r_s = 0.525$, $p = 0.01$; anxiety: $r_s = 0.526$, $p = 0.01$; depression: $r_s = 0.689$, $p = 0.01$). The stigma enacted-subscale was also positively correlated with all the DASS-21 scales (stress: $r_s = 0.440$, $p = 0.01$; anxiety: $r_s = 0.244$, $p = 0.05$; depression: $r_s = 0.465$, $p = 0.01$), indicating that higher levels of stigma are associated with higher levels of stress, anxiety and depression, with self (felt) stigma having a stronger correlation than enacted.

Regression

Hierarchical block regression models were conducted; see Tables 4 to 6 for the results of this analysis.

Insert Table 4 here please

Insert Table 5 here please

Insert Table 6 here please

Regression model predicting stress.

Demographic and clinical variables made up less than 5% of the variance at predicting the DASS-21 stress scores, whereas, social support added a further 15% of the variance, which was increased by a further 25% of variance to a total of 45% variance (41% adjusted R^2 variance) when the stigma self-subscale and enacted-subscale were added to the model. The overall model at step four was significant ($F = 9.671, p = < 0.001$).

Social support was identified as a statistically significant predictor for stress at step three of the model ($\beta = -.408, p = < 0.001$), however this was no longer statistically significant when stigma (self-subscale and enacted-subscale) entered the model at step four.

At step four only the self-subscale of stigma was a significant independent predictor of stress ($\beta = .434, p < 0.001$).

Regression model predicting anxiety.

For the DASS-21 anxiety scores, a different pattern emerged. Demographic and clinical variables accounted for less than 5% of the variance, social support added a further 17% of the variance, and the stigma self-subscale and enacted-subscale added a further 15% of variance for a model total of 37% of the variance (31% adjusted R^2 variance). The final model was significant ($F = 6.709, p < 0.001$).

Social support was identified as a significant predictor for anxiety at step three of the model ($\beta = -.434, p < 0.001$). It remained a significant independent predictor at step four ($\beta = -.260, p < 0.05$), alongside self-stigma ($\beta = .485, p < 0.001$).

Regression model predicting depression.

The model predicting the greatest variance was for the DASS-21 depression scores. Demographics and clinical variables accounted for less than 4% of the variance, social support added a further 22% of variance and stigma total score added a further 30% of variance, resulting in a total model variance of 56% (52% adjusted R^2 variance). The final model was significant ($F = 14.917, p < 0.001$).

Social support was identified as a statistically significant predictor of depression at step three of the model ($\beta = -.490, p < 0.001$). It just remained a significant independent predictor at step four ($\beta = -.192, p < 0.05$), alongside the self-subscale of the stigma measure ($\beta = .603, p < 0.001$).

Discussion

The present study investigated the impact of social support and perceived stigma in contributing to psychological distress in individuals with MND. Statistically significant correlations were found between social support, stigma (total score, self-subscale and

enacted-subscale) and all the outcome variables, highlighting the significance of these relationships. Hierarchical regression analyses revealed that enacted stigma was not an independent predictor in any of the models and social support did not remain a significant independent predictor for stress when stigma entered the model. Moreover, felt stigma was a more powerful predictor than social support in each of the models.

This study is the first, to the best of the authors knowledge, to investigate social support as a predictor of psychological distress (conceptualised as stress, anxiety and depression) in individuals with a diagnosis of MND controlling for demographics and physical functioning. The results of this study suggest that individuals with higher levels of perceived social support have lower levels of psychological distress. This indicates that having access to adequate social support can be a protective factor for avoiding psychological distress for individuals with a diagnosis of MND. These findings are consistent with one of the most dominant theories in social support research; the stress buffering model (Barrera, 1986; Cohen & Wills, 1985; Cutrona & Russell, 1990; Thoits, 1986) which is an extension of the Lazarus (1966) and Lazarus and Folkman's (1984) general stress and coping theory (Lakey & Orehek, 2011). Applied in the context of this study, this theory suggests that social support would act as a buffer between MND related stress and psychological distress and thus reduce distress.

By having adequate support from friends, family and significant others with regards to emotional and practical matters, some of the difficulties that individuals with MND experience could be improved. These difficulties include having to develop and adjust their adaptation strategies as the condition progresses, as well as functional changes which can impact the individual's ability to complete tasks or engage socially due to experiences of changing identity, social status and social relationships (Mistry & Simpson, 2013). Having

high levels of support in place to help manage these transitions and experiences can be beneficial to the individual's levels of psychological distress.

Turning to stigma, previous research had identified stigma as a significant predictor of quality of life in individuals with a diagnosis of MND (van der Beek et al., 2013), however this study did not use an exclusively MND sample, and the focus was on quality of life, rather than psychological distress. Therefore, this research is the first, to the best of the author's knowledge, to identify the predictive nature of stigma for psychological distress, in a sample which was exclusively individuals with a diagnosis of MND.

The correlational findings indicate that individuals with higher levels of stigma (both felt and enacted) have higher levels of psychological distress. However, in the regression analyses, only the self-subscale (measuring felt stigma) was a significant independent predictor of psychological distress. These results partially support the findings by van der Beek et al., (2013) who found that both the enacted-subscale and self-subscale components were independent significant predictors of quality of life. However, the self-subscale was identified as a stronger independent predictor than the enacted-subscale component. This suggests that felt or internalised beliefs about being stigmatised are more influential on psychological distress than actual experiences of discrimination (enacted). Felt stigma was a stronger predictor than enacted stigma and came out independently when predicting the variance for each of the models. Interestingly, Scambler and Hopkins (1986) found that 90% of their participants with a diagnosis of epilepsy had experienced felt stigma, whereas only a third of these had experienced enacted stigma. They suggested that this discrepancy between levels of felt and enacted stigma may be attributed to the public being more informed and tolerant of the condition, than expected by academics and individuals with epilepsy. This was also theorised by Furnham and Lane (1984) with regards to individuals experiencing deafness.

Strengths and Limitations

The majority of participants were male (55.8%), meaning this sample differed from more specific estimates of prevalence rates which suggest that MND is 54% higher in males than females (Alonso, Logroscino, Jick & Hernán, 2009). The mean age of this sample was 59 years, which is younger than the average age of onset of between 60 and 65 years identified in past research (Talbot & Marsden, 2008). The mean time since symptom onset for this sample was 4.75 years (median 3.5 years), which can be considered a long duration, as the median survival rate following symptom onset has been reported as between 2 to 4 years (Chiò et al., 2009). Therefore, the time since symptom onset for this sample was longer than would be expected. Moreover, 10% of the participants in this sample had a symptom duration time that exceeded 10 years, in line with the figures suggested by Chiò et al., 2009 which suggests that 10-20% of individuals survive past 10 years following symptom onset. However, as this study was conducted online, this may bias the sample towards younger people. Also, most participants in this study (55%) had higher levels of physical functioning, indicating that they were able to function independently despite the symptom durations reported. This higher rate of physical functioning may relate to the online nature of the study design, in that those who chose to participate in the study were those who had more ability to access it.

This study used a cross-sectional design which has known limitations such as the difficulty in making causal inferences and only gathering data at a single time-point (Levin, 2006). The results suggest that having high levels of stigma correlate with high levels of depression, however the direction of this relationship cannot be assumed. It is possible that this is a bi-directional relationship and that having high levels of depression could cause high levels of felt stigma as an individual may be more susceptible to internalising negative perceptions about their condition. Previous research has identified a bi-directional

relationship between stigma and depression in a sample of children affected by HIV/ AIDS, whereby higher levels of depression predicted higher levels of perceived stigma which then predicted higher levels of enacted stigma. This suggested a vicious cycle as higher levels of enacted stigma also directly predicted subsequent higher levels of depression (Chi, Li, Zhao & Zhao, 2014). Therefore, understanding whether a similar relationship exists for individuals with MND would be beneficial. Alternatively, it could be that high levels of felt stigma cause higher levels of depression due to negative consequences of feeling stigmatised. In order to improve this and allow causal inferences to be made, longitudinal studies need to be conducted, using more complex statistical models such as regressions that predict the outcomes at subsequent timepoints. Using statistical models that unpack the mechanisms of these relationships, such as identifying mediating and moderating effects would also add to our understanding.

Sample size is a limitation in this study, as only a relatively small sample size was achieved, however, challenges were encountered trying to access this population due to the relative rarity of the condition. An *a priori* power calculation had been conducted prior to recruitment commencing, which estimated that a minimum of 92 participants would be required to detect a medium effect size (0.15) with 80% power, and 5-8 predictors. However, the final models (with 6 predictors) actually found large effects (explaining 30-52% of the variance in the outcomes) and post-hoc calculations suggest the power achieved was over 99% for the whole models. Nonetheless, social support may have remained a significant predictor in the stress model if a larger sample size had been achieved.

Another issue that was encountered during this study was the recruitment of participants. Due to the low prevalence rates of MND, an online survey method was used to recruit participants internationally. This was a successful method to increase participant numbers, however, this relied on organisations and support services in individual countries to

advertise and share the study information in order to recruit participants and certain countries such as the USA were hard to reach in this respect. Only one local organisation (The Les Turner Foundation) in the USA shared this information, resulting in only a small proportion (3%) of participants from this country. Nonetheless this method of recruitment was successful in various other countries and a sufficient sample size was gained.

A further limitation to the study was that participants were only recruited if they were able to understand English well enough to complete the study measures, so the results may not be generalisable across cultures and ethnicities. Only nationality, not ethnicity, was recorded as part of the demographic questionnaire and the majority of these (87%) reported an English-speaking nationality. Also, participants were accessed online, biasing the sample towards those who had the means and ability to access the internet. This could result in some populations without internet access being underrepresented or neglected from the current sample.

Furthermore, the levels of stigma were quite low in this sample, and most of the sample had 'normal' levels of stress, anxiety and depression. A review of psychosocial aspects of MND (McLeod & Clarke 2007) identified varying prevalence rates of depression and anxiety within MND populations, these ranged from 0% to 50% for depression (defined as moderate to severe) and 11% to 26% for anxiety. Therefore, given the high variability, it is unclear whether the current sample may be considered representative. However, the present findings may not be applicable to samples with higher levels of stigma and psychological distress.

Clinical Implications

The findings from this study provide implications for clinical practice, as they highlight the importance of perceived social support and stigma in predicting psychological distress for individuals with a diagnosis of MND. This suggests that if psychological distress

is identified in an individual with MND, then it may be beneficial to use social and psychological understandings and interventions aimed at reducing felt stigma and enhancing social support.

Interventions aimed at targeting stigma often operate on several levels; intrapersonal, interpersonal, organisational/ institutional, community and governmental/ structural (Heijnders & Van Der Meij, 2006). Systematic reviews have identified that the most effective interventions are aimed at the intrapersonal, interpersonal and community levels (Heijnders & Van Der Meij, 2006; Rao et al., 2019). Effective intervention strategies for reducing stigma for conditions such as HIV, mental health diagnoses and leprosy include education (Arole, Premkumar, Arole & Maury, 2002; Brown, 2009; Ngoc, Weiss & Trung, 2016; Patalay et al., 2017), counselling (Jürgensen, Sandøy, Michelo & Fylkesnes, 2013; Lusli et al., 2016), cognitive behavioural therapy (Corrigan & Calabrese, 2005, Hall & Tarrier, 2003) social marketing (Henderson et al., 2012), drama therapy (Orkibi, Bar & Eliakim, 2014) and social support groups (Demissie, Getahun & Lindtjørn, 2003; Lyon & Woodward, 2003; Thurman, Jarabi & Rice, 2012) along with combinations of these different approaches (contact, counselling and education: Uys et al., 2009; education, contact and social marketing: Pinfold, Thornicroft, Huxley & Farmer, 2005; social support and counselling: Fawzi et al., 2012). Moreover, research suggests that mindfulness is positively associated with stigma resistance in individuals with a psychiatric diagnosis (Chan, Lee & Mak, 2018) suggesting that mindfulness-based interventions may be beneficial in reducing stigma. Mindfulness-based interventions have also been identified as effective in reducing psychological distress for individuals with a diagnosis of MND (Pagnini et al., 2015; Pagnini et al., 2017).

The concept of psycho-emotional disablism (Simpson & Thomas, 2014) is a useful conceptual framework for understanding how society can impose limitations and barriers to

an individual, making it appear that they lack these abilities. The term ‘disablism’ was defined by Thomas (2007) as “a form of social oppression involving the social imposition of restrictions of activity on people with impairments and the socially engendered undermining of their psycho-emotional wellbeing” (p.73). By taking this approach towards an individual’s levels of impairment, as opposed to a pathologising, individualising approach, clinical psychologists can aim to effect societal change to combat stigma by advocating for the individuals they work with, with the aim of working towards the individuals ‘real world’ goals and objectives. This framework aims to effect change at a community/ structural level and when used in combination with the above intrapersonal, interpersonal and community interventions could begin address the negative influence that stigma can have on the psychological well-being of individuals with a diagnosis of MND.

Social support interventions may also be beneficial to reduce psychological distress, however there is a lack of evidence regarding their effectiveness, particularly for individuals with MND. A meta-analysis conducted by Pfeiffer, Heisler, Piette, Rogers and Valenstein (2011) found that social support interventions, such as peer support, have been found to help reduce levels of depression in mixed samples of individuals, however, this did not include a specific MND sample. Therefore, more research is needed investigating social interventions which may increase social support and decrease stigma. This can help develop the area of psychological support for the management of psychological distress on offer to individuals with a diagnosis of MND, which has been previously identified as an area in need of development (McLeod & Clarke, 2007; Oh & Kim, 2017).

Future Research

As social support and stigma both significantly predict psychological distress in individuals with MND it may be advantageous to explore the relationship between these variables further. The stress buffering model (Barrera, 1986; Cohen & Wills, 1985; Cutrona

& Russell, 1990; Thoits, 1986) suggests that social support acts as a buffer between MND related stress and psychological distress. Therefore, research to assess whether social support acts as a buffer between stigma (or other MND-related stressors) and psychological distress would be beneficial to determine the relationship between these variables. The data collected from this research could be reanalysed to test the stress buffering hypothesis.

Furthermore, research using more complex models to identify the direction of the relationship between stigma and depression for individuals with MND could be beneficial, as there is a lack of evidence in this area, yet previous research has identified this in other populations such as children affected by HIV/AIDS (Chi et al., 2014).

Conclusion

The findings of the present study indicate that social support and stigma are both significantly correlated with psychological distress in individuals with a diagnosis of MND. Regression analyses revealed that enacted stigma was not an independent predictor in any of the models and social support did not remain a significant independent predictor for stress when stigma entered the model. Moreover, felt stigma was a significant independent predictor in all the models and was a more powerful predictor than social support in each of the models. These findings can have a beneficial impact upon individuals with a diagnosis of MND as they highlight the importance of considering social and psychological factors when psychological distress has been identified. This can then inform management strategies and psycho-social interventions to help reduce the levels of psychological distress experienced by this population. Further research investigating the relationship between the variables of social support and stigma may also be beneficial.

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Table 1:*Demographic information of participants*

Variable	Sample
Age (years)	
Mean (<i>SD</i>)	59.14 (10.60)
Range	36 – 83
Gender	
Male (%)	43 (55.8)
Female (%)	34 (44.2)
Nationality (%)	
Australian	8 (5.8)
British	12 (8.7)
Canadian	14 (10.1)
Caucasian	1 (0.7)
Dutch	1 (0.7)
English	2 (1.4)
German	1 (0.7)
Indian	1 (0.7)
Irish	3 (2.2)
New Zealander	18 (13)
Northern Irish	1 (0.7)
NZ European	3 (2.2)
Scottish	1 (0.7)
South African	4 (2.9)

Swedish	1 (0.7)
UK	2 (1.4)
USA	2 (1.4)
USA Canadian	1 (0.7)
White	1 (0.7)
Relationship Status (%)	
Single	3 (2.2)
Cohabiting	3 (2.2)
Married/ civil partnership	55 (39.9)
Divorced	9 (6.5)
Widowed	4 (2.9)
Other	3 (2.2)
Time since symptom onset (years)	
Mean (<i>SD</i>)	4.75 (4.41)
Median	3.5
Range	0.6 – 22
Time since diagnosis (years)	
Mean (<i>SD</i>)	3.53 (3.92)
Median	2.4
Range	0.1 to 21
Carer assistance required to complete survey (%)	
Yes	7 (9.1)
No	70 (90.9)

Table 2: *Descriptive statistics for all study measures*

Variable	Mean	Standard Deviation	Cronbach's alpha
DASS-21 Stress	7.03	4.59	.86
DASS-21 Anxiety	5.32	3.85	.75
DASS-21 Depression	7.19	5.22	.91
SA-ALSFRS-R	29.00	8.62	.82
MSPSS Total	66.47	15.66	.94
MSPSS Significant Other	24.10	5.68	.94
MSPSS Family	22.16	6.16	.90
MSPSS Friends	20.21	6.27	.91
SSCI Total	32.83	16.12	.92
SSCI Self-Subscale	22.48	10.36	.90
SSCI Enacted-Subscale	10.09	7.09	.87

Note: **DASS-21** = Depression, Anxiety and Stress Scale; **SA-ALSFRS-R** = Self-Administered

Amyotrophic Lateral Sclerosis Functional Rating Scale Revised; **MSPSS** = Multidimensional Scale of Perceived Social Support; **SSCI** = Stigma Scale for Chronic Illness.

Table 3: *Correlation matrix for variables displaying Spearman's correlation coefficients*

Variable	Age	Gender	SA-ALSFRS-R	MSPSS Total	SSCI Total	SSCI Self	SSCI Enacted	DASS-21 Stress	DASS-21 Anxiety	DASS-21 Depression
Age	1.000	.090	-.031	.198	-.134	-.140	-.179	-.175	-.026	-.052
Gender		1.000	-.165	-.097	.028	.014	-.016	.114	-.067	-.070
SA-ALSFRS-R			1.000	.255*	-.402**	-.395**	-.232*	-.153	-.209	-.180
MSPSS Total				1.000	-.483**	-.483**	-.433**	-.385**	-.399**	-.437**
SSCI Total					1.000	.929**	.801**	.538**	.447**	.660**
SSCI Self						1.000	.586**	.525**	.526**	.689**
SSCI Enacted							1.000	.440**	.244*	.465**
DASS-21 Stress								1.000	.627**	.787**
DASS-21 Anxiety									1.000	.588**
DASS-21 Depression										1.000

* $p < 0.05$; ** $p < 0.01$

Table 4: *Summary of hierarchical multiple regression analysis model for DASS-21 Stress*

	Standardised Beta Coefficient	R²	Adjusted R²	R Square Change	F
Step 1		.034	.008	.034	1.306
Age	-.167				
Gender	.095				
Step 2		.049	.010	.014	1.243
Age	-.166				
Gender	.075				
SA-ALSFRS-R	-.122				
Step 3		.203*	.159*	.155**	4.596*
Age	-.075				
Gender	.059				
SA-ALSFRS-R	-.064				
MSPSS Total	-.408**				
Step 4		.453**	.406**	.250**	9.671**
Age	-.046				
Gender	.090				
SA-ALSFRS-R	-.083				
MSPSS Total	-.107				
SSCI Self-Subscale	.434**				
SSCI Enacted-Subscale	.239				

* $p < 0.05$; ** $p < 0.001$

Table 5: *Summary of hierarchical multiple regression analysis model for DASS-21 Anxiety*

	Standardised Beta Coefficient	R²	Adjusted R²	R Square Change	F
Step 1		.002	-.025	.002	0.86
Age	-.001				
Gender	.048				
Step 2		.046	.006	.043	1.163
Age	.004				
Gender	.013				
SA-ALSFRS-R	-.211				
Step 3		.220**	.177**	.175**	5.091**
Age	.100				
Gender	-.004				
SA-ALSFRS-R	-.149				
MSPSS Total	-.434**				
Step 4		.365**	.311**	.145**	6.709**
Age	.109				
Gender	.010				
SA-ALSFRS-R	-.024				
MSPSS Total	-.260*				
SSCI Self-Subscale	.485**				
SSCI Enacted-Subscale	-.069				

* $p < 0.05$; ** $p \leq 0.001$

Table 6: *Summary of hierarchical multiple regression analysis model for DASS-21 Depression*

	Standardised Beta Coefficient	R²	Adjusted R²	R Square Change	F
Step 1		.001	-.026	.001	.053
Age	-.031				
Gender	.019				
Step 2		.039	-.001	.037	.984
Age	-.028				
Gender	-.052				
SA-ALSFRS-R	-.196				
Step 3		.262**	.221**	.223**	6.400**
Age	.080				
Gender	-.071				
SA-ALSFRS-R	-.126				
MSPSS Total	-.490**				
Step 4		.561**	.524**	.299**	14.917**
Age	.103				
Gender	-.044				
SA-ALSFRS-R	.049				
MSPSS Total	-.192*				
SSCI Self-Subscale	.603**				
SSCI Enacted-Subscale	.084				

* $p < 0.05$; ** $p < 0.001$

Appendix 2-A: Author Guidelines



BJHP AUTHOR GUIDELINES

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1. SUBMISSION

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Section 3: Critical Appraisal

Psychological perspectives on stigma and social support for individuals with multiple sclerosis and motor neurone disease

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Word count (exc. title page, references and appendix): 3,973

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This section of the thesis aims to provide a critical appraisal of aspects of the whole thesis. Firstly, I will provide an overview of the findings from the systematic literature review and empirical research paper. I will discuss the similarities and differences between multiple sclerosis (MS) and motor neurone disease (MND) and the relevance of these in relation to social support and stigma. I will then discuss the issues which I encountered during the recruitment process for the empirical paper and the conceptual framework of disability used to guide the terminology applied to both papers. I will then discuss some personal reflections on why I chose this research topic and what I learned from this process. Finally, I will make recommendations for future research.

Main Findings

Systematic Literature Review

The review focused on psychological correlates of social support for individuals with a diagnosis of MS. The definition for a psychological variable adopted for the systematic review was “potentially modifiable factors relating to the individual’s attitudes, thoughts, feelings, and behaviours that would be relevant and possible to address in a psychological intervention” (Dennison, Ross-Morris & Chalder, 2009, p.142).

A systematic review identified 21 studies that assessed the relationship between social support and psychological variables which met the above definition. The psychological variables assessed in the 21 studies were anger, anxiety, coping, depression, hope, loneliness, mental aspects of health-related quality of life, mental health status, post-partum emotional distress and psychological adjustment. The studies included in the review reported correlational and regression analyses and all the studies theorised social support as a predictor in their research.

Statistically significant correlations were identified for all the above psychological variables apart from coping. Higher levels of social support were correlated with lower levels of anger, anxiety, depression, loneliness and psychological adjustment. Higher levels of social support were also correlated with higher levels of hope, mental aspects of health-related quality of life, mental health status, postpartum emotional distress and self-esteem.

In regressions, greater social support was also shown to be a statistically significant predictor of less anxiety, anger, depression and loneliness. Furthermore, greater social support was also found to be a significant predictor of better mental aspects of health-related quality of life, postpartum emotional distress and self-esteem. The correlations with the largest effect size were between social support and depression and mental composite of quality of life. The variable with the largest number of studies and significant results was depression, followed by mental aspects of health-related quality of life. Mental aspects of health-related quality of life and depression also had some of the highest quality assessment ratings, strengthening these findings.

Empirical Research Paper

The empirical research paper aimed to answer the research question: does a lack of social support and perceived stigma contribute to psychological distress in individuals with motor neurone disease (MND)? Seventy-seven participants were recruited through various international organisations supporting individuals with MND (see Appendix 3-1 for list of organisations who shared the study details) and through social media. Participants were required to complete an online survey consisting of the study measures. To control for potential confounders, measures were included to assess demographic variables and physical functioning, along with the two variables of interest (social support and stigma). Finally, one

measure was used to assess the outcome of psychological distress (measuring stress, anxiety and depression).

Statistically significant relationships were found between the main predictors of stigma and social support and all three outcome variables. Higher levels of stigma were correlated with higher levels of stress, anxiety and depression and higher levels of social support were correlated with lower levels of stress, anxiety and depression.

Greater social support was identified as a statistically significant predictor for lower stress, anxiety and depression, when controlling for demographics and physical functioning and remained significant when stigma entered the model, for anxiety and depression, but not for stress. Greater felt stigma was identified as a statistically significant predictor for higher levels of stress, anxiety and depression and all the final models were significant. Therefore, stigma and social support may be important considerations for ameliorating psychological distress in people with MND.

The Relationship Between MS, MND, Social Support and Stigma

MS and MND are both neurodegenerative conditions, eliciting neurological symptoms in the individual which progressively worsen over time (Chaudhuri, 2013; Gordon, 2011). There are also similarities in terms of the types of symptoms that individuals experience as both conditions affect muscle movement and mobility, damage the brain and spinal cord and cause scarring or hardening around nerve cells (Amor & van Noort, 2012; Leigh et al., 2003; Howard & Orrell, 2002; Brown & Al-Chalabi, 2017). However, there are substantial differences between the conditions, mainly with regards to the course of progression and outcome. MS is not classified as a life-limiting illness, as life expectancy is only shortened by a few months (Rolak, 2003), however, this can increase to a shortened life expectancy of 5-10 years for 30% of individuals with relapsing-remitting MS (Raffel, Wakerley & Nicholas, 2016). In contrast to this, MND is considered a life-limiting illness

and median survival times following symptom onset are generally two to four years, with only 10%-20% of individuals surviving past 10 years (Chiò et al., 2009). Furthermore, the progression of functional impairment also differs between the two conditions. In MND the progression of functional impairment is typically rapid and relentless (Brown & Al-Chalabi, 2017); conversely, MS initially presents with a relapsing-remitting presentation of functional impairment in 90% of cases (Raffel et al., 2016). This relapsing-remitting presentation is highly variable and symptoms can fluctuate significantly (Rolak, 2003). Although this is not present in all cases, the progressive nature of other forms of MS - primary-progressive; secondary-progressive; and progressive-relapsing - occur over a much longer time period than that of MND (Lorscheider et al., 2016; Rocca et al., 2017; Salemi et al., 2013).

These differences are relevant when conducting research on social support and stigma. As the progression of the condition in MS typically fluctuates in the initial stages, it would be expected that the need for social support would also fluctuate. Research investigating the role of social support in MS have found that individuals with MS recruit more social support when their levels of impairment are higher (Rommer, König & Zettl, 2016) but that they perceive less social support when their functional impairment is higher (O'Brien, 1993), i.e. despite the higher level they do not feel it meets their needs at that time. It could also be hypothesised that individuals with MS would have higher levels of overall social support than those with MND, as when the symptoms of MS are in remission, then access to social relationships and opportunities to socialise are more available. Conversely, for individuals with MND the functional impairment presents a persistent and rapid decline, suggesting that their access to social support and relationships would decrease as their condition progressed. Moreover, research has identified that individuals with MND experience higher levels of stigma than those with MS (Molina, Choi, Cella & Rao, 2013). Further research has also identified that for individuals with MND, stigma is a significant

predictor of social withdrawal which may partially mediate the effects of functional impairment (Schlüter, Tennant, Mills, Diggle & Young, 2018).

Empirical Paper Recruitment Appraisal

I encountered challenges during the recruitment process for the empirical paper. One challenge related to accessing participants, as MND is a relatively rare condition, affecting only approximately 5,000 individuals in the UK (National Institute for Health and Care Excellence, 2015). Worms (2001) estimated a prevalence rate for North America and Europe of around two per 100,000 of the population. Therefore, it was difficult to recruit a large sample for this study although an *a priori* power calculation (using G*Power software based on 5-8 predictors, presuming a medium effect size of 0.15) suggested between 92 and 109 participants were required (80% power, $\alpha = .05$). To reach this target, I recruited internationally to maximise my chances of gaining a sufficient sample. I therefore used Qualtrics software to allow the survey to be conducted on an online platform, accessible to an international audience, however this assumed that participants would have internet access. Additionally, there are limitations to this method, such as biasing the recruitment towards a younger and more functionally able audience (Age UK, 2018; Topolovec-Vranic & Natarajan, 2016). I recruited a fairly young sample which had a mean age below that of the average age of onset for MND. Moreover, the majority of the sample I recruited had 'normal' levels of depression, anxiety and stress and had mild to moderate levels of physical impairment. Research suggests that younger people are more likely to participate in research which is advertised through social media and the internet (Topolovec-Vranic & Natarajan, 2016). Furthermore, Age UK (2018) recently reported that 36% of individuals aged 65 and older do not currently use the internet and that those with mobility difficulties have a decreased likelihood of using the internet. Therefore, using an online survey and advertising

my study online and via social media, could have contributed to the recruitment of a younger and more functionally able sample. On balance I feel this was an appropriate method, as despite the limitations, I was able to reach participants that I may not have accessed through alternative methods. However, future research may benefit from employing alternative recruitment strategies such as conducting face-to-face surveys at clinics, hospitals and care homes as well as utilising postal surveys in order to enhance the representation within the sample.

Another challenge that I was presented with was gaining access to US participants. I was able to access successfully participants from a range of countries, such as Australia, Canada, New Zealand and the UK through contact with organisations supporting individuals in these countries (see Appendix 3-A for list of organisations and how they shared the study information). However, the main organisation supporting individuals with MND in the US would not share my study advert or details with their members, as the research was not being conducted by themselves, so they were unable to promote this. This meant that I was not able to access a large pool of potential participants, which impacted the final sample size. I tried alternative methods of recruiting from this country, by 'tagging' the individual chapters of this organisation into my study advert posts and I also contacted other, smaller organisations to see if they would be willing to share the details. Only one of these other organisations offered to share my study details via different methods such as group meetings and through staff appointments. However, despite this organisation expressing their willingness to share the information, only two of the participants recruited declared their nationality as 'USA'. Therefore, if I were to undertake this process again, I would consider establishing links with organisations in the US a priority from the outset of the process. Perhaps consulting them during the establishment of the research would have provided them with a role and incentive to collaborate with the recruitment process.

Interestingly, the country from which I recruited the greatest number of participants was New Zealand with a total of 21 participants reporting New Zealand or New Zealand European as their nationality.

Another issue that I encountered during recruitment was the time taken to recruit participants. I kept the survey online for as long as was practically possible and the overall duration for data collection was around 18 months. This was a long duration considering the timescale for completion of a doctoral thesis and was only achieved as, during the data collection period, I had a period of maternity leave. I would therefore advocate that researchers conducting research within this field take into consideration the amount of time required to recruit a sufficient sample size from this population.

Conceptual Framework of Disability

As this thesis sits within the clinical health psychology field, it felt important to consider the stance which I took with regards to concepts such as disability and the terminology used throughout. I assumed a critical position towards the concept of disability and considered an alternative conceptual framework in which to view this to guide the write up of this thesis. This was in an attempt to assume a socio-psychological position, as opposed to a diagnostic, pathologising approach which medical literature often adopts. Literature relating to MS and MND often refers to the 'levels of disability' (Amor, & van Noort, 2012; Andersen et al., 2012; Bromberg, 2015; Gulick, 1992; Kraft, Freal & Coryell, 1986; Talbot & Marsden, 2008; Walsh & Walsh, 1989) that the condition brings. However, this view can bring damaging assumptions with regards to an individual's impairment and alternative views of the concept of 'disability' have been proposed to overcome the unhelpful narrative that can be associated with the term. Simpson and Thomas (2014) argued that the concept of 'structural and/or psycho-emotional disablism' is a useful conceptual framework

for understanding how society can impose limitations and barriers to an individual, making it appear that they lack these abilities. The term ‘disablism’ was defined by Thomas (2007) as “a form of social oppression involving the social imposition of restrictions of activity on people with impairments and the socially engendered undermining of their psycho-emotional wellbeing” (p.73) and an individual’s ‘level of disability’ was viewed in the context of this concept throughout the literature review and empirical paper. Therefore, the term ‘disability’ was replaced with the term impairment when considering physical limitations imposed by the condition, as I would argue that disability is a product of societal limitations as well as the physical limitations.

Another adjustment made to terminology was reflected in the empirical paper. ‘Emotional lability’ is a diagnostic term that refers to involuntary changes in mood where strong feelings and emotions can occur, such as uncontrollable laughing or crying (Palmieri et al., 2009) and was used throughout the literature. For the purpose of this thesis, this term was referred to as involuntary changes in mood as this is a more descriptive and meaningful way of expressing this experience for an individual. This is also more in line with psychological concepts regarding emotional distress, fitting with a psychological formulation approach as opposed to a psychiatric diagnostic system (Johnstone & Dallos, 2014).

Personal Reflections on the Research Process

My reasons for embarking on this research project related to my own research interests in the area of neurodegenerative conditions, particularly MND. My interest in MND stems from my own personal experiences of wanting to understand more about this condition and the psychological impacts of such a diagnosis, as my sister-in-law had personal experiences of this condition. Her father was diagnosed with MND and she was his main carer until he passed away. I have been privileged enough to gain an insight into some of the challenges that the condition presented to her father and their family members through her

sharing her experiences. I therefore wanted to explore this further and research some of the psychological impacts of this condition, as these aspects are often overlooked (McLeod, 2007; Oh & Kim, 2017). Through discussions with my sister-in-law regarding the psychological challenges that can be presented, stigma was discussed, as the perceptions and misconceptions of others often generated feelings of distress and could make practical tasks more difficult to manage. The importance of family and friends was also discussed as a protective factor and through this, the concept of loss of social support as the condition progressed was identified. The loss of social support can compound stigma, increasing psychological distress for the individual and their family, therefore I wanted to investigate whether this experience was shared by others experiencing this condition.

After consulting the literature base, I identified a gap in this area as little or no research had been conducted assessing the influence of stigma and social support on psychological distress for people with MND. Social support has been identified as a significant predictor of quality of life and depression in this population (Matuz, Birbaumer, Hautzinger & Kübler, 2010; Matuz, Birbaumer, Hautzinger & Kübler, 2015), but this used a very small sample size. Furthermore, anxiety and stress had not been investigated. Stigma has been identified as a significant predictor of quality of life for individuals with a neuromuscular condition (van der Beek, Bos, Middel and Wynia, 2013). However, individuals diagnosed with MND only made up 9% of the total number of participants in this study and distress had not been investigated.

Having chosen a topic for my research paper, I then had to identify an appropriate and complementary systematic literature review. I chose to focus on social support as more research had been conducted in this area, providing a larger set of studies to review. I then chose to examine the relationship between social support and psychological variables, as this complemented the concept of psychological distress being investigated in the empirical

paper. However, there were insufficient studies investigating the relationship between social support and psychological variables in people with MND. I therefore considered an alternative neurodegenerative condition which was more thoroughly researched than MND, MS. Two previous reviews have been conducted in this area, which have focused on depression and MS (Arnett, Barwick & Beeney, 2008) and psychosocial correlates of adjustment in MS (Dennison et al., 2009). There was some overlap between these and the current review, as perceived social support was included as a psychosocial factor within the Dennison et al. (2009) review and as a correlate of depression in the Arnett et al. (2007) review. However, these reviews were conducted at least 10 years ago, and social support was not the focus of these reviews. Furthermore, the way in which the Dennison et al. (2009) review was theorised differs from the current review, as they were seeking predictors of adjustment outcomes, whereas my review was seeking research investigating social support and its relationship with other psychological variables. Therefore, the current review aimed expand on these, by identifying more recent research and by assessing the specific concept of perceived social support.

Throughout the process of conducting this research, I gained a deeper appreciation of the challenges and difficulties experienced by individuals with a diagnosis of MND and their family members. This impacted me personally in relation to my sister-in-law as, although I had previously recognised the impact this may have had, I had not appreciated the magnitude of this. I have gained a greater respect for how she coped with these challenges and the resilience she has developed in order to manage these experiences. This was compounded when I received emails from both individuals with a diagnosis of MND and by a family member who had recently lost her partner to the condition. They shared their experiences with me of how distressing their journey had been from receiving the diagnosis, managing as the condition progressed, and ultimately the end of life stages. Hearing these stories provided

a profound contextual awareness to the statistical data that I had collected, by providing the human element that is often overlooked when conducting quantitative research.

Future Research

Through the process of critically appraising this thesis, recommendations for future research were identified. One recommendation relating to my personal reflections would be for more qualitative research to be conducted, exploring the stories of both individuals with MND and their family members/ carers. Specifically, qualitative research investigating the experiences of felt and enacted stigma, and the role which social support plays in relation to this. This type of research could enrich the findings from this thesis by developing a contextual narrative in which the present results can be compared and viewed. This could also inform future quantitative research, as through qualitatively exploring the relationships between stigma, social support and psychological distress, the mechanisms through which these relationships operate could be identified then tested through statistical methods.

During the systematic review process, I also identified that some qualitative research has been conducted on psychological variables such as coping (Dehghani, Neyeri & Ebadi, 2017); adaptation (Dilorenzo, Becker-Feigeles, Halper & Picone, 2008), well-being (Hamed, Tariah & Hawamdeh, 2012) and the individuals experience of living with MS (Barton, Magilvy & Quinn, 1994). However, no reviews have been conducted in the area of psychological variables, to the best of the author's knowledge, and this topic may benefit from future research.

After consulting the literature base to identify potential reasons for differences between research participation uptake between different countries, I discovered a dearth of research in this area. It would therefore be beneficial for research to be conducted which compares the attitudes towards participation in health research for different countries and

cultures. This could enhance the recruitment and participation process for future research, as this could identify countries in which attitudes towards participation is less favourable then work could be undertaken to establish the reasons behind this. Moreover, more resources and methods of engagement could be applied to these countries/ cultures to enhance recruitment and establish more representative findings.

During the recruitment process and through my personal reflections it has become apparent that research into the impacts of stigma and a lack of social support in relation to psychological distress for the family members and carers of individuals with a diagnosis of MND is required. I was contacted through twitter by a partner of an individual with MND asking if families and children could participate in this research as the impact that the condition had on their three children was of deep concern. They added that the fallout of this condition was quite unique, and they were pleased to see interest shown in this most neglected aspect of MND. Therefore, the psychological distress resulting from the impact of this condition is not only felt by the individual with MND, but their family members and carers too. Hence, future research exploring this further is required to establish whether this experience is shared by other families and to what extent the psychological well-being of family members is impacted by stigma and lack of social support. The results could be used to advocate for recognition of this impact by support services, so family members and carers can receive the support required to address these needs and interventions can be advised accordingly. Additionally, stigma interventions aimed at a community/ structural levels as outlined in the empirical paper of this thesis would also benefit family members and carers alongside the individual with MND.

Conclusions

I feel that I met the research aims and that the two papers complemented each other, as although they focused on two separate conditions, these both sit within the neurodegenerative discipline and the findings from each paper identified similar relationships between social support and variables representing psychological well-being/ distress. There was coherence in terms of the predictor of interest (social support) and the inclusion of stigma in the empirical paper was also fitting with the concept of social factors and identity, which can include how individuals perceive themselves and the support they receive from others.

Despite the issues encountered during the recruitment process of the empirical paper and the sample size being limited, this did not impact upon the identification of medium and large effect sizes in the results. This suggests that a sufficient sample size was gained, and the overall process of recruitment was a success regardless of the challenges faced.

The results of this thesis might be helpful in identifying clinical implications to raise awareness of the potential psychological consequences resulting from a lack of social support and experiencing felt stigma. The findings might be beneficial to guide intervention planning to ensure that an individual's psychological needs are recognised, addressed and appropriate interventions offered. Recommendations for future research were also identified which could enhance the current literature base available and further explore these relationships to identify the mechanisms through which these relationships operate. Moreover, qualitative research is advocated to explore the individual experiences of both individuals with MND and their family members/ carers to enrich the quantitative research already established.

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Appendix 3-A: List of Organisations

Organisation	Country	Support Provided
ALS Canada	Canada	Shared survey link on social media (Facebook and Twitter).
ALS/MND Alliance	International organisation	Sent study details to worldwide associations asking them to share with their members. Shared survey link on Twitter.
ALS Society Quebec	Canada	Shared survey link on their social media and in their province. Also shared with ALS Canada
Lancaster Centre for Ageing Research	UK	Shared study details on social media.
Les Turner Foundation	USA	Shared with their staff to cascade to members and through their support groups.
Minds and Movement	UK	Shared study details on social media.

MNDA	UK	Advertised study on the research page of their website and in their newsletter.
MNDA Australia	Australia	Shared the study details on social media.
MNDA New Zealand (NZ)	New Zealand	Shared with NZ MND Registry and shared on website and social media.
MNDA Scotland	Scotland	Shared on Twitter and with MND clinical specialists to cascade.
MNDA South Africa	South Africa	Shared study details with their members and in their newsletter.
NZ MND Registry	New Zealand	Shared study details with all the members on their research database.

Section 4: Ethics Section

**Does a lack of social support and perceived stigma
contribute to psychological distress in individuals
with motor neurone disease?**

Natalie Leigh*

Doctorate in Clinical Psychology, Lancaster University

Word count (exc. appendices): 5,693

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FHMREC Application Form

1

Faculty of Health and Medicine Research Ethics Committee (FHMREC)

Lancaster University

Application for Ethical Approval for **Research****for additional advice on completing this form, hover cursor over 'guidance'.****Guidance on completing this form is also available as a word document**

Title of Project: Does perceived stigma influence psychological distress in individuals with motor neurone disease?

Name of applicant/researcher: Natalie Leigh

ACP ID number (if applicable)*:

Funding source (if applicable)

Grant code (if applicable):

***If your project has *not* been costed on ACP, you will also need to complete the Governance Checklist [\[link\]](#).**

Commented [guidance1]: In completing your application:

- Be specific and thorough in the information you provide in the application form. You may use short sections from your research protocol, cutting and pasting as relevant.
- Make sure that the *application form* includes all the important points of information about the study so that committee members can evaluate your submission thoroughly from the application form itself. This may mean repeating (or cutting and pasting) elements of your protocol.
- Be sure that information provided is consistent between your application, research protocol, and supporting materials. Repeating information is likely to be necessary.
- Organise the information clearly.
- If you have different phases or components to your research, clearly present and address issues for each component (using headings, etc.) so that it is easy to understand what you plan to do.
- For projects involving human participants, and being reviewed by the full committee: a lead reviewer is appointed from the FHMREC who will take the lead on reviewing your application. The lead reviewer may contact you prior to the meeting if any clarification of your application is needed.

Commented [guidance 2]:

Make your title short and descriptive so that people can easily identify the main topic of the research; you do not need extensive detail. The title of your project does not need to be the same as the title you propose to use for your publication (e.g. your thesis).

Type of study

- ☐ Involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants. **Complete sections one, two and four of this form**
- ☒ Includes *direct* involvement by human subjects. **Complete sections one, three and four of this form**

2

SECTION ONE

1. Appointment/position held by applicant and Division within FHM Student on Doctorate in Clinical Psychology Programme

2. Contact information for applicant:

E-mail: n.leigh@lancaster.ac.uk **Telephone:** 07508 375657 (please give a number on which you can be contacted at short notice)

Address: Doctorate in Clinical Psychology, Furness Building, Lancaster University, Lancaster, LA1 4YG

3. Names and appointments of all members of the research team (including degree where applicable)

Dr Fiona Eccles - Research Supervisor - Lecturer, Division of Health Research, Lancaster University

Dr Jane Simpson - Field Supervisor - Director of Education, Division of Health Research, Lancaster University

3. If this is a student project, please indicate what type of project by marking the relevant box/deleting as appropriate: (please note that UG and taught masters projects should complete FHMREC form UG-tPG, following the procedures set out on the [FHMREC website](#))

PG Diploma ☐ Masters by research ☐ PhD Thesis ☐ PhD Pall. Care ☐

PhD Pub. Health ☐ PhD Org. Health & Well Being ☐ PhD Mental Health ☐ MD ☐

DClinPsy SRP ☐ [if SRP Service Evaluation, please also indicate here: ☐] DClinPsy Thesis ☒

4. Project supervisor(s), if different from applicant: Dr Fiona Eccles - Research Supervisor and

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Dr Jane Simpson - Field Supervisor

5. Appointment held by supervisor(s) and institution(s) where based (if applicable): Dr Fiona Eccles - Lecturer, Division of Health Research, Lancaster University

Dr Jane Simpson - Director of Education, Division of Health Research, Lancaster University

SECTION TWO

Complete this section if your project involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants

1. Anticipated project dates (month and year)

Start date:

End date:

2. Please state the aims and objectives of the project (no more than 150 words, in lay-person's language):

Commented [guidance 3]:

These dates should indicate when recruitment will begin, (taking into account the timescale of the ethical approval process) and when funding ends or your thesis will be submitted.

Commented [guidance 4]:

This summary should concisely but clearly tell the reader (in simple terms and in a way which would be understandable to a general audience) what you are broadly planning to do in your study.

Data Management

For additional guidance on data management, please go to [Research Data Management](#) webpage, or email the RDM support email: rdm@lancaster.ac.uk

3. Please describe briefly the data or records to be studied, or the evaluation to be undertaken.

4a. How will any data or records be obtained?

4b. Will you be gathering data from websites, discussion forums and on-line 'chat-rooms' ☐ no

4c. If yes, where relevant has permission / agreement been secured from the website moderator? ☐ no

4d. If you are only using those sites that are open access and do not require registration, have you made your intentions clear to other site users? ☐ no

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4e. If no, please give your reasons

5. What plans are in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc)? Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with the Data Protection Act 1998.

6a. Is the secondary data you will be using in the public domain?

6b. If NO, please indicate the original purpose for which the data was collected, and comment on whether consent was gathered for additional later use of the data.

Please answer the following question *only* if you have not completed a Data Management Plan for an external funder

7a. How will you share and preserve the data underpinning your publications for at least 10 years e.g. PURE?

7b. Are there any restrictions on sharing your data?

8. Confidentiality and Anonymity

a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications?

b. How will the confidentiality and anonymity of participants who provided the original data be maintained?

9. What are the plans for dissemination of findings from the research?

Commented [guidance5]: Data storage: non-audio and non-video data. State clearly where and what format your data will be stored.

Timescales: The standard guidance we provide to people about length of time for retaining data is 10 years. This is a requirement. Your study may have a rationale for retaining data longer and for various intended purposes, but if so, please explain. For example, some data may be specifically collected with intent to be added to a formal databank (quantitative or qualitative), or there may be plans for secondary data analysis that is anticipated from early in the design of the project. Where electronic data is to be stored for longer than the recommended period, we recommend storing data on University storage. If data is collected or stored by own devices they need to be encrypted. For data sharing with external partners we recommend using Box.

Data Stewardship: Please state who will have guardianship of the stored data (and if you are a student, who will be responsible for storing/deleting your data once you have completed your course). For DMP purposes: how will the data be documented and described i.e. how will you make others understand your data. We recommend writing a description/abstract, adding a Readme file, using meaningful file names and version numbers.

Location: If your data is stored centrally or will be accessible to others, you should note in your application who will have access to the data.

Data security: Data stored on all portable devices (eg laptops) should be encrypted as well as pass word protected; data stored on the University server does not, however, need to be encrypted. If you are based and work predominantly away from the University, give consideration to how you will store the data securely as you undertake your research, and how it will be securely transferred to the LU campus for long term storage.

Commented [guidance6]: Example 1: Supporting data will be provided in an electronic format on the journal website, with unrestricted access post-publication. Data will also be deposited in Lancaster University's institutional data repository and made freely available with an appropriate data license. Lancaster University uses Pure as the data repository which will hold, manage, preserve and provide access to datasets produced by Lancaster University research.

Example 2: All relevant files with documentation will be offered to the UK Data Archive as per the standard ESRC procedures. If the UK Data Archive will not accept the offered data, it will be stored in Lancaster University's data repository (via Pure) where it will be preserved according to Lancaster University's Data Policy for a minimum of 10 years.

For DMP purposes: You can use PURE to share and preserve your data with LU data archive or deposit data with an external trustee...

Commented [guidance7]: Example: Due to the small sample size, even after full anonymization there is a small risk that participants can be identified. Therefore, supporting data will only be shared on request with genuine researchers. Access will be granted on a case by case basis by the Faculty of Health and Medicine.

Commented [guidance 8]: Dissemination covers a wide range of activities including (but not limited to) reports, academic submissions (such as theses and dissertations), study summaries, and publications:
 • Please state who will see the data - ie research team members.
 • If you are a student, be sure to include your academic paper (such as dissertation or thesis) as a form of dissemination.
 • Phrasing regarding publication should reflect that you may pursue submission for publication, but you cannot guarantee that the dissemination will include publication. For example, you may write "Results of the research may be submitted for publication in an academic/professional journal."

5

10. What other ethical considerations (if any), not previously noted on this application, do you think there are in the proposed study? How will these issues be addressed?

SECTION THREE

Complete this section if your project includes *direct* involvement by human subjects

1. Summary of research protocol in lay terms (indicative maximum length 150 words):

This study aims to investigate whether an individual's level of perceived stigma influences psychological distress for individuals with motor neurone disease (MND). This requires approximately 100 English speaking participants who will be accessed internationally via an online survey. The survey requires completion of questionnaires which assess: perceived experience of stigmatisation, psychological distress (depression, anxiety, stress and life satisfaction) and symptom severity. The results will then be analysed to discover if those participants who experience high levels of stigmatisation experience a higher level of psychological distress. The research will also investigate if the strength of this relationship is stronger than that of other factors, such as demographics (age, gender, nationality and relationship status), clinical factors (time since symptom onset, time since diagnosis and symptom severity) and another psychological factor (social support). It is anticipated that stigma will have a significant effect over and above other predictors of psychological distress.

Commented [guidance 9]:

The summary of the protocol should concisely but clearly tell the reader (in simple terms and in a way which would be understandable to a general audience) what you are broadly planning to do in your study. A helpful format may include a sentence or two about the background/ "problem" the research is addressing, why it is important, followed by a description of the basic design and target population. Think of it as a snapshot of your study. Again, it is fine to cut and paste relevant information from your research protocol.

2. Anticipated project dates (month and year only)

Start date: 09/2017

End date: 05/2018

Commented [guidance 10]:

These dates should indicate when recruitment will begin, (taking into account the timescale of the ethical approval process) and when funding ends or your thesis will be submitted.

Data Collection and Management

For additional guidance on data management, please go to [Research Data Management webpage](#), or email the RDM support email: rdm@lancaster.ac.uk

3. Please describe the sample of participants to be studied (including maximum & minimum number, age, gender):

Commented [guidance 11]:

- Indicate any inclusion/exclusion criteria.
- Indicate if there will be specific language restrictions, and if so, why. For example, it is common, especially for students, that studies are restricted to English speaking participants due to limited funding for research conduct (and thus translation/interpretation), limited time for collecting or analysing data in other languages, or lack of applicability of language diversity due to the specific population being targeted. These are just examples but demonstrate that lack of group inclusion needs to be thoughtfully explained.
- Where particular groupings are being used, give details of how you will determine that a participant belongs to this group. For example if you intend to include participants from ethnic minority backgrounds in your study, you will need to explain how the ethnicity of potential participants will be identified.

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Participants will include men and women aged 18 or over who have a diagnosis of MND/ amyotrophic lateral sclerosis (ALS)/ Lou Gehrig disease. Inclusion criteria includes having English to a level to understand the questionnaires and having a diagnosis of MND or equivalent (amyotrophic lateral sclerosis; ALS or Lou Gehrig disease), identified through self-report.

An a priori power calculation (using G*Power based on 5-8 predictors, presuming a medium effect size suggests approximately 100 participants will be required (80% power, alpha = .05).

4. How will participants be recruited and from where? Be as specific as possible. Ensure that you provide the *full versions* of all recruitment materials you intend to use with this application (eg adverts, flyers, posters).

Participants will be recruited internationally mainly from English speaking countries, although participants who speak English who are based in other countries will be eligible too. Participants will be accessed through advertising on social media, through charities and support services advertising the study on their websites/ social media accounts and through their newsletters, using the study advert. I also aim to speak to charities and support services directly to see if they will allow me to advertise at their venues using the study advert as well as through electronic means. This will be aimed at charities and support services both in the UK and in other countries such as the USA, Canada, Australia and New Zealand. However, people who speak English and live in other countries will also be eligible to take part.

5. Briefly describe your data collection and analysis methods, and the rationale for their use.

Questionnaires will be used to gain data via an online survey method, this will also include self-report questions for demographic and clinical data.

These questionnaires will assess levels of stigma (SSCI; Rao et al., 2009) and levels of psychological distress: DASS-21 (Lovibond & Lovibond, 1995). Measures will also be used to assess levels of social support (MSPSS; Zimet et al., 1988) and symptom severity (Self-Administered Amyotrophic Lateral Sclerosis Functional Rating Scale; SA-ALSFRS-R; Cedarbaum et al., 1999; Montes et al., 2006). Data will also be collected via self-report method for demographic variables such as age, gender, nationality and relationship status and clinical variables such as time since symptom onset and time since diagnosis. The demographic information being collected is for descriptive purposes only, as this will be used to describe the sample rather than being used to test a specific hypothesis. The demographic information is required in order to provide relevant and contextual details of the sample population to inform discussion of the results and for comparison with previous research.

Once the survey has begun then the information provided will be anonymous meaning that no personal identifiable information will be held. The study findings will be produced into a report which may be published, however no personal identifiable information will be included in the report.

Commented [guidance 12]:

- Exactly what steps will you take to recruit participants?
- How will they find out about the study?
- How will they indicate that they want to participate?
- Will all volunteering participants be included or do you need to take into consideration turning some away?
- If you plan to use different recruitment procedures for different participant groups, clearly indicate this and outline each set of procedures under a subheading so that it is easy to see which procedures apply to which group.
- If you will be using a test to determine participants' eligibility, please indicate how you will apply the test and how you will handle communicating with people who do not meet the eligibility criteria.

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If a participant wishes to access a copy of the results of the study then their personal details will be held long enough to send out this information, then it will be destroyed and this information will be kept confidential throughout the process. The data collected for this study will be stored securely and only the researchers conducting this study will have access to this data:

o The files on the computer will be encrypted (that is no-one other than the researcher will be able to access them) and the computer itself password protected.

o Lancaster University will keep your anonymised data for a period of 10 years after the study has finished.

The method of analysis which is to be used is a hierarchical block regression model. Predictors will be entered in four blocks: Demographics (age, gender, nationality and relationship status), clinical variables (symptom severity, time since symptom onset and time since diagnosis), a psychological variable (social support) and stigma. These blocks will predict the psychological distress outcome variables (depression, anxiety and stress). This will be completed using SPSS software. This is to understand if scores of stigma predict scores on measures of psychological distress as it is hypothesised that stigma will have a significant effect over and above other predictors of psychological distress. The inclusion of other predictors allows for comparison between these and stigma to identify which is the strongest predictor for psychological distress in this population.

6. What plan is in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc.)? Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with the Data Protection Act 1998.

All data will be gained through Qualtrics software and stored securely electronically (e.g. on the university server or on Box). This data will not be personally identifiable and will be kept by Lancaster University DClinPsy course for 10 years at the end of which period it will be destroyed by DClinPsy Programme Staff under the direction of the research supervisor (Fiona Eccles).

7. Will audio or video recording take place? ☒ no ☐ audio ☐ video

a. Please confirm that portable devices (laptop, USB drive etc) will be encrypted where they are used for identifiable data. If it is not possible to encrypt your portable devices, please comment on the steps you will take to protect the data. N/A

b. What arrangements have been made for audio/video data storage? At what point in the research will tapes/digital recordings/files be destroyed?

N/A

Commented [guidance 13]:

Data storage: non-audio and non-video data. State clearly where and what format your data will be stored.

Timescales: The standard guidance we provide to people about length of time for retaining data is 10 years. This is not a requirement but a *general recommendation*. Your study may have a rationale for retaining data longer and for various intended purposes, but if so, please explain. For example, some data may be specifically collected with intent to be added to a formal databank (quantitative or qualitative), or there may be plans for secondary data analysis that is anticipated from early in the design of the project. Where electronic data is to be stored for longer than the recommended period, it should only be kept on Lancaster University servers, and not on portable or home devices.

For DMP purposes: how will the data be documented and described i.e. how will you make others understand your data. We recommend writing a description/abstract, adding a README file, using meaningful file names and version numbers.

Data Stewardship: Please state who will have guardianship of the stored data (and if you are a student, who will be responsible for storing/deleting your data once you have completed your course).

Location: If your data is stored centrally or will be accessible to others, you should note in your application who will have access to the data.

Data security: Data stored on all portable devices (eg laptops) should be encrypted as well as password protected; data stored on the University server does not, however, need to be encrypted.

If you are based and work predominantly away from the University, give consideration to how you will store the data securely as you undertake your research, and how it will be securely transferred to the LU campus for long term storage.

Commented [guidance14]: Transporting audio/video data:

you should state that if you store any identifiable data (audio recordings, participant contact details etc) on portable devices such as a memory stick or laptop you will use encryption. Password protection alone is not sufficient for identifiable data. Information on encryption is available from ISS <http://www.lancs.ac.uk/iss/security/encryptionoptions/> and their service desk is also able to assist.

If your portable device cannot be encrypted, you must confirm that any identifiable data (including recordings of participants' voices) will be deleted from the recorder as quickly as possible (eg when they have been transferred to a secure medium, such as a password protected & encrypted PC) and state that the device will be stored securely in the meantime.

For DMP purposes: state explicitly about the anticipated volume of data collected. This has implications for storage planning and cost calculations especially if data are images or videos. Contact ISS if data will reach 100GB.

Commented [guidance 15]:

Storage. Audio and video data is considered more sensitive than most written data because of its capacity to threaten confidentiality more directly. There are, however, no fixed deadlines, and recordings such as oral histories may be kept in perpetuity. With audio data that does not need to be kept for the long term, it is common to erase/destroy the recording once it has been transcribed and checked. However, we suggest that you retain the recordings until your work has been examined and/or published, in case you need to check the original recordings for any reason. For video, it may depend on the types of analyses proposed for the study. There may be good reason to keep the data longer, but the key in completing this section of the application form is to be explicit about timescales for storage, and the reasons for your timescale should be clearly indicated and explained.

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Please answer the following questions *only* if you have not completed a Data Management Plan for an external funder

8a. How will you share and preserve the data underpinning your publications for at least 10 years e.g. PURE?

The raw data will not be publicly available and it will not be uploaded to PURE. The raw data will be held electronically on the secure Lancaster University server by the DCLinPsy Programme for 10 years following submission of the data.

8b. Are there any restrictions on sharing your data?

The raw data will not be shared publicly.

9. Consent

a. Will you take all necessary steps to obtain the voluntary and informed consent of the prospective participant(s) or, in the case of individual(s) not capable of giving informed consent, the permission of a legally authorised representative in accordance with applicable law? yes

b. Detail the procedure you will use for obtaining consent?

There is a consent form that will be shown to individuals prior to beginning the online survey. Once they have read this and clicked a link to say they have read the participant information sheet, understand the points and agree to consent then this will be recorded as providing their consent.

10. What discomfort (including psychological eg distressing or sensitive topics), inconvenience or danger could be caused by participation in the project? Please indicate plans to address these potential risks. State the timescales within which participants may withdraw from the study, noting your reasons.

There is no discomfort expected for the participants, however due to discussing their experiences of stigma and well-being there could be the potential to evoke distress and as such if they experience any distress following participation they will be encouraged to contact the resources provided at the end of the participant information and debrief sheets.

In terms of gaining valid consent, all participants will be aged 18 or over and will be provided with an information sheet which they will have the option of downloading and can ask questions by contacting the researcher prior to completing the survey and providing their consent. They will be unable to withdraw their data once the survey has begun as all data is not identifiable at this stage, this is outlined in the consent form

11. What potential risks may exist for the researcher(s)? Please indicate plans to address such risks (for example, noting the support available to you; counselling considerations arising from the sensitive or distressing nature of the research/topic; details of the lone worker plan you will follow, and the steps you will take).

Commented [guidance16]: Example 1: Supporting data will be provided in an electronic format on the journal website, with unrestricted access post-publication. Data will also be deposited in Lancaster University's institutional data repository and made freely available with an appropriate data license. Lancaster University uses Pure as the data repository which will hold, manage, preserve and provide access to datasets produced by Lancaster University research.

Example 2: All relevant files with documentation will be offered to the UK Data Archive as per the standard ESRC procedures. If the UK Data Archive will not accept the offered data, it will be stored in Lancaster University's data repository (via Pure) where it will be preserved according to Lancaster University's Data Policy for a minimum of 10 years.

For DMP purposes: You can use PURE to share and preserve your data with LU data archive or deposit data with an external trusted repository. Consider if anonymised data can be shared openly or if it needs access restrictions e.g. share only on request.

Commented [guidance17]: Example: Due to the small sample size, even after full anonymization there is a small risk that participants can be identified. Therefore, supporting data will only be shared on request with genuine researchers. Access will be granted on a case by case basis by the Faculty of Health and Medicine.

Commented [guidance 18]:

Be specific:

- When in the research process will consent be taken?
- By whom? Where? How?
- Will there be non-handwritten forms of consent being used in the research, and if so, what will they be and why will they be used?
- For those using anonymous surveys for data collection, no consent form will be used because that would compromise anonymity. However, a cover sheet or some type of introduction should clearly inform participants that by completing the survey they are providing consent for the use of the data for research. The cover sheet or introduction may also remind participants of other aspects of wh...

Commented [guidance 19]:

Be as thorough as possible in anticipating potential sources of discomfort:

- Provide a plan for addressing the discomfort that may arise during the conduct of the research and discomfort that may develop following the conduct of the research, potentially as a consequence of participation in the research. We suggest you include possible sources of support in the Participant Information Sheet. You may also consider providing a debriefing sheet.

Commented [guidance 20]: •Time limits for withdrawing from the study: when writing about how you might address risk, please avoid the phrase "participants may withdraw at any time" because withdrawal for most studies is time limited. For example, once you have published your data, withdrawal is clearly not possible in the true sense. You may want to consider a reasonable time period for withdrawal following data collection, depending on the type of study you are doing, for example:

- 1. If you are collecting interview data and will be conducting simultaneous data collection and analysis, it may be reasonable to give participants a 2 week period following the interview to ...

Commented [guidance 21]:

Clearly indicate potential risks and plans for reducing/addressing them:

- If referring to a Lone Worker Policy, please indicate which policy you are following (from which agency or source). However, you should not simply state that you will be following a given policy: you also need to outline the main elements of it that apply to you. You may wish to use the University's Lone Worker Guidance (field work) as the basis of this section:

<http://www.lancs.ac.uk/depts/safety/files/fieldguidance.pdf> ...

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There are no risks to the researchers as the study is conducted online. The topic area is not a distressing one, although the client population could cause some distress to the devastating nature of the disease. Support is available to the researcher from research and field supervisors along with a clinical tutor.

12. Whilst we do not generally expect direct benefits to participants as a result of this research, please state here any that result from completion of the study.

There will be no direct benefit to the participants for taking part in the study.

13. Details of any incentives/payments (including out-of-pocket expenses) made to participants:

As the survey is being completed online there will be no out-of-pocket expenses. There are no incentives/ payments being offered to participants.

14. Confidentiality and Anonymity

a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications? yes

b. Please include details of how the confidentiality and anonymity of participants will be ensured, and the limits to confidentiality.

Once the survey has begun then the information provided will be anonymous meaning that no personal identifiable information will be held.

15. If relevant, describe the involvement of your target participant group in the *design and conduct* of your research.

I have gained service-user input from two experts by experience, who have contributed to the design and development of the research. One of these experts by experience was a carer for an individual with MND and the other was a member of the Lancaster University Public Involvement Network, with an interest in neurological conditions. This input included feedback regarding the materials and advertising along with the selection of appropriate measures for the target population and the duration of the survey. I have approached the Motor Neurone Disease Association and discussed the study with them. They were willing to advertise for experts by experience to contribute to the design of the study, however this was not possible within the timeframe available. They are also willing to advertise the advert once ethical approval has been granted in order to assist in the recruitment of participants for the study.

16. What are the plans for dissemination of findings from the research? If you are a student, include here your thesis.

Commented [guidance 22]:

In most situations, it is important to acknowledge that there may not be (or will not be) direct benefit to participants. Potential phrasing is "There may be no direct benefit to participation in this study." However, it may be appropriate to add that people may find it a positive experience to participate in the research because of (specify reasons that are relevant for your research). You may also include benefits to society. This will vary from project to project.

Commented [guidance 23]:

If you are intending to use incentives/payments, keep in mind that they should be modest so as not to suggest coercion of the participants. If you are reimbursing for travel, please indicate the financial limit of the reimbursement.

Commented [guidance 24]:

Be specific, and address the requested information completely:

- Be sure to address the rationale for both data collection and the process of analysis.
- Limits of Confidentiality: You should include information about the situations in which you would need to break confidentiality (harm to self or others) and what you will do should you need to break confidentiality.
- Ensure that you differentiate between anonymity and confidentiality. EG where you use direct quotes from participants in publications you cannot be said to have maintained confidentiality, though it is possible to assure anonymity.
- Confidentiality of participation: where you are carrying out interviews on work premises, perhaps with the agreement of a participant's line manager, clarify here how you will ensure confidentiality of participants; or note that you will make it clear that confidentiality cannot be fully ensured in these circumstances. You should also consider and address confidentiality for participants who are from a small community, and whose comments may be easily attributed.
- State your intention to anonymise the project participants, and how this will be done.
- If you are having your data transcribed, please note clearly who will be doing this. If the transcriber will be external to LU research team, please ensure participants are informed, and that a confidentiality agreement has been signed by the transcriber. This agreement should be submitted with your application.
- If you intend to interview participants at a distance using video-conferencing it is recommended that you use the University's WebEx system, as this is the most secure. If this is not possible, and you need to use Skype you should:

Commented [guidance 25]:

This section relates to how you have included your *target participation group* in the design and conduct of your research. We encourage you to include user involvement in your research whenever possible. For example, if your target participant group have provided consultation on recruitment procedures or have provided feedback on drafts of surveys that will be used in the study, this should be described in this section.

Commented [guidance 26]:

Dissemination covers a wide range of activities including (but not limited to) reports, academic submissions (such as theses and dissertations), study summaries, and publications:

- Please state who will see the data - ie research team members. If your data collection methods mean that others will be present (eg interpreters), please note what additional requirements have been considered in terms of consent.
- If you are a student, be sure to include your academic paper (such as dissertation or thesis) as a form of dissemination.
- Phrasing regarding publication should reflect that you may pursue submission for publication, but you cannot guarantee that the dissemination will include publication. For example, you may write:

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Following completion, the study findings will be submitted as my DClinPsy thesis and will be presented to peers and staff at Lancaster University. This presentation will be available on the University's website for the public and trainee clinical psychologists to access. There could be the opportunity to publish the findings in the Motor Neurone Disease Association newsletter and on their website, along with any other support services that have been approached. The findings may also be put forward for publication in a peer reviewed journal and/ or presented at conferences.

17. What particular ethical considerations, not previously noted on this application, do you think there are in the proposed study? Are there any matters about which you wish to seek guidance from the FHMREC?

Commented [guidance 27]:

It is rare that studies have no ethical considerations at all. Try to be thorough and thoughtful when considering this question. You should not try to invent issues, and at the same time, do not assume that by noting a problem you are hurting your application. This section provides an opportunity for you to demonstrate to the committee that you have a substantial and clear understanding of the potential ethical issues, and that you have given thought to how to address them (even if they may not be able to be addressed perfectly).

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SECTION FOUR: signature

Applicant electronic signature: Date

Student applicants: please tick to confirm that you have discussed this application with your supervisor, and that they are happy for the application to proceed to ethical review ☐

Project Supervisor name (if applicable): Date application discussed

Commented [guidance 28]:

If you are a student, make sure that you have discussed the project and the application with your supervisor. Build in enough time in your preparation schedule for your supervisor to properly review your application and give you comments before you give it to him/her for signing.

Submission Guidance

1. Submit your FHMREC application by email to Diane Hopkins (d.hopkins@lancaster.ac.uk) as two separate documents:
 - i. **FHMREC application form.**
Before submitting, ensure all guidance comments are hidden by going into 'Review' in the menu above then choosing *show markup>balloons>show all revisions in line*.
 - ii. **Supporting materials.**
Collate the following materials for your study, if relevant, into a single word document:
 - a. Your full research proposal (background, literature review, methodology/methods, ethical considerations).
 - b. Advertising materials (posters, e-mails)
 - c. Letters/emails of invitation to participate
 - d. Participant information sheets
 - e. Consent forms
 - f. Questionnaires, surveys, demographic sheets
 - g. Interview schedules, interview question guides, focus group scripts
 - h. Debriefing sheets, resource lists

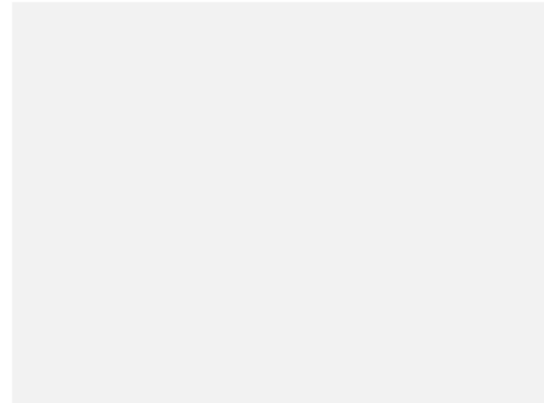
Please note that you DO NOT need to submit pre-existing measures or handbooks which support your work, but which cannot be amended following ethical review. These should simply be referred to in your application form.

Commented [guidance 29]: We recommend that you use and amend the sample information sheet and consent form on the FHM Research Ethics Committee website as the basis for these documents. Ensure that you use the most recent relevant logo on your participant materials. Consider any special requirements that your intended research participants may have, and endeavour to accommodate these. For example if your participants include people with a learning disability, your information sheet and consent form should include pictorial representations of the information you are trying to communicate (Makaton). For more information go to <http://www.makaton.org/>

2. Submission deadlines:
 - i. Projects including direct involvement of human subjects [section 3 of the form was completed]. The *electronic* version of your application should be submitted to Diane Hopkins by the committee deadline date. Committee meeting dates and application submission dates are listed on the [FHMREC website](#). Prior to the FHMREC meeting you may be contacted by the lead reviewer for further clarification of your application. Please ensure you are available to attend the committee meeting (either in person or via telephone) on the day that your application is considered, if required to do so.
 - ii. The following projects will normally be dealt with via chair's action, and may be submitted at any time. [Section 3 of the form has *not* been completed, and is not required]. Those involving:

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- a. existing documents/data only;
 - b. the evaluation of an existing project with no direct contact with human participants;
 - c. service evaluations.
3. You must submit this application from your Lancaster University email address, and copy your supervisor in to the email in which you submit this application



Thesis Protocol

Title: Does perceived stigma influence psychological distress in individuals with motor neurone disease?

Name of applicant/supervisors/affiliations/version number:

Applicant: Natalie Leigh, Trainee Clinical Psychologist, Lancashire Care NHS Foundation Trust

Field Supervisor: Dr Jane Simpson, Director of Education, Division of Health Research, Lancaster University

Research Supervisor: Dr Fiona Eccles, Lecturer, Division of Health Research, Lancaster University

Version number: One

Introduction:

Motor neurone disease (MND), also referred to as amyotrophic lateral sclerosis (ALS) and Lou Gehrig disease in the United States of America, is a fatal neurodegenerative disease (Pagnini, Philips, Bosma, Reece & Langer, 2015). This condition affects the motor neurons in the brain and spinal cord and alters an individual's ability to control voluntarily their muscle movements, leading to paralysis, swallowing difficulties, respiratory failure and ultimately, and only within a few years from diagnosis, death (Dib, 2003).

Psychological distress is an important factor when considering individuals with a diagnosis of MND, not only in and of itself, but also because individuals with a higher level of well-being may have a lower risk of mortality than those experiencing distress, even when disease severity and length of illness is considered (McDonald, Wiedenfeld, Hillel, Carpenter & Walter, 1994). Therefore, an individual's level of psychological distress can be viewed as a protective factor in terms of the impact of the diagnosis and exploring this further is also important given its contribution to overall quality of life.

Due to the devastating nature of the condition, individuals with a diagnosis of MND are likely to experience psychological distress and decreased well-being (Pagnini et al., 2015). Although a number of factors undoubtedly contribute to this, recent research has suggested that stigma may influence an individual's sense of well-being in a negative manner in different populations living with physical health conditions including those experiencing lung, head and neck cancer (Lebel et al., 2011), individuals with epilepsy and migraines (Aydemir, Özkara, Ünsal & Canbeyli, 2011), individuals diagnosed with obesity (Carr & Friedman, 2005), individuals with Parkinson's disease (Maffoni, Giardini, Pierobon, Ferrazzoli & Frazzitta, 2017) and individuals living with a diagnosis of HIV (Sanjuán, Molero, Fuster & Nouvilas, 2012).

The term stigma was originally defined by Goffman (1963) as "an undesired differentness" and this has since been expanded to include two concepts of stigma; felt and enacted (Scambler & Hopkins, 1986). 'Felt stigma' refers to a feeling of shame about being different and feeling that discrimination for this difference will occur, whereas, 'enacted stigma' refers to actual experience of this discrimination.

The influence of stigma as a predictor of quality of life in individuals diagnosed with neuromuscular diseases, including MND, has been studied by Van Der Beek, Bos, Middel and Wynia (2013). This study found that stigma was a major predictor of poorer quality of life, with 'felt stigma' being a stronger predictor compared with 'enacted stigma'. However, this study was focused on quality of life, rather than psychological distress in particular, and individuals diagnosed with MND only made up 9% of the total number of participants. Due to the fatality of MND in comparison to the other neuromuscular diseases included within the study (muscle disorder, junction disorder and peripheral nerve disorder), it seems important to study this population independently.

Another important predictor to consider alongside stigma is social support. Social support has been identified as an important factor in relation to stigma in a variety of conditions, including depression, where it has been reported that most research has found a negative association between the two factors (Mickelson, 2001). Both social support and stigma have been identified as predictors of quality of life for individuals with epilepsy (Whatley, Dilorio & Yeager, 2010) and these factors have also been identified as predictors which contribute significantly to post-traumatic growth for individuals with a diagnosis of HIV (Zeligman, Barden & Hagedorn, 2016). It

therefore feels important to also assess how the social aspects of a condition can affect psychological distress in individuals with MND.

Rationale:

This research aims to discover if there is a relationship between perceived stigma and psychological distress for people with a diagnosis of MND. Also, discovering whether the strength of this relationship is stronger than that of other factors will be considered, such as demographics (age, gender, nationality and relationship status), clinical factors (time since symptom onset, time since diagnosis and symptom severity) and the psychological factor of social support. It is believed that stigma will have a significant effect over and above other predictors of psychological distress. If results support this, then this will highlight the importance of considering perceived stigma when assessing psychological distress in an individual diagnosed with MND. This will provide evidence suggesting that providing interventions aimed at reducing perceptions of stigma would be beneficial and aims to address the causes of stigma and identify the levels of stigmatisation individuals with MND experience to promote awareness and challenge this socially.

Research Question:

The research question posed for this study is: Does perceived stigma influence psychological distress in individuals with motor neurone disease?

Research Design:

This will be a quantitative study using outcome measure scores for perceived stigma: Stigma Scale for Chronic Illnesses (SSCI; Rao et al., 2009) and levels of psychological distress: Depression, Anxiety and Stress Scale (DASS-21; Lovibond & Lovibond, 1995). Demographic predictors will also be included in the analysis (age, gender, nationality and relationship status), along with clinical predictors including symptom severity (Self-Administered Amyotrophic Lateral Sclerosis Functional Rating Scale; SA-ALSFRS-R; Cedarbaum et al., 1999; Montes et al., 2006), time since symptom onset and time since diagnosis. A psychological predictor will also be included which measures social support (Multidimensional Scale of Perceived Social Support; MSPSS; Zimet, Dahlem, & Farley, 1988). The hypothesis will be analysed using a hierarchical block regression model to consider other potential predictors alongside the predictor of interest (stigma).

Participants:

Participants will include men and women aged 18 or over who have a self-reported diagnosis of MND or equivalent (e.g. ALS/ Lou Gehrig disease). Inclusion criteria includes having English to a level to understand the questionnaires and having a diagnosis of MND or equivalent (e.g. ALS/ Lou Gehrig disease; confirmed through self-report).

An a priori power calculation (using G*Power based on 5-8 predictors, presuming a medium effect size suggests approximately 100 participants will be required (80% power, alpha = .05).

Materials:

Stationery will be required for advertising materials (see Appendix A).

A participant information sheet (see Appendix B), consent form (see Appendix C) and debrief sheet (see Appendix D) will be required.

An electronic version of each of the questionnaires, including the self-report measures, will be required to input to the online survey (see Appendix E – K)

Qualtrics software will be required to deliver the survey online.

Recruitment:

Participants will be recruited internationally by advertising the survey through various methods including through charities/ support services/ social media networks in the UK and internationally. This is due to the limited number of individuals with a diagnosis of MND in the UK alone which is estimated at 5,000 adults at any one time (Motor Neurone Disease Association; MNDA, 2017). A Qualtrics survey will then be used to collect the data.

Procedure:

Questionnaires will be used to gain data via an online survey method, this will also include self-report questions for demographic and clinical data.

These questionnaires will assess levels of stigma (SSCI; Rao et al., 2009) and levels of psychological distress (DASS-21; Lovibond & Lovibond, 1995). Measures will also be used to assess levels of social support (MSPSS; Zimet et al., 1988) and symptom severity (Self-Administered Amyotrophic Lateral Sclerosis Functional Rating Scale; SA-ALSFRS-R; Cedarbaum et al., 1999; Montes et al., 2006). Data will also be collected via self-report method for demographic variables such as age, gender, nationality and relationship status and clinical variables such as time since symptom onset and time since diagnosis.

Once the survey has begun then the information provided will be anonymous meaning that no personal identifiable information will be held. The study findings will be produced into a report which may be published, however no personal identifiable information will be included in the report. If a participant wishes to access a copy of the results of the study then their personal details will be held long enough to send out this information, then it will be destroyed and this information will be kept confidential throughout the process. The data collected for this study will be stored securely and only the researchers conducting this study will have access to these data:

- The files on the computer will be encrypted (that is no-one other than the researchers will be able to access them) and the computer itself password protected.
- Lancaster University will keep the anonymised data for a period of 10 years after the study has finished.

Data Analysis:

The method of analysis which is to be used is a hierarchical block regression model. Predictors will be entered in four blocks: Demographics (age, gender, nationality and relationship status), clinical variables (symptom severity, time since symptom onset and time since diagnosis), the psychological variable (social support) and stigma. These blocks will predict the outcome variable of psychological distress (depression, anxiety and stress). This will be completed using SPSS software. This is to understand if scores of stigma predict scores on measures of psychological distress as it is hypothesised that stigma will have a significant effect over and above other predictors of psychological distress. The inclusion of other predictors allows for comparison between these and stigma to identify which is the strongest predictor for psychological distress in this population.

Practical Issues:

A potential issue is regarding recruitment as there is a low prevalence rate of MND in the UK alone. To address this the survey will be completed online allowing international access to the survey and advertising will be targeted internationally to expand the recruitment pool. This will be aimed at English speaking countries and at individuals who speak English that live in other countries, however, the eligibility criteria for the study requires the ability to speak English proficiently in order to understand the questionnaires. Advertising will be aimed at support services and through social media to access as many participants as possible.

Moreover, due to the nature of MND, the physical functioning of participants may be reduced meaning that depending upon the participant's stage of the disease, their ability to enrol themselves in the study or manually complete the online surveys may be an issue. If this is the case and the participant does not have the physical capability or assistive technology to complete the survey, but does wish to take part then it may be possible to have a carer complete this alongside the participant based upon the participant's communicated responses.

Dissemination:

Following completion, the study findings will be submitted as my DClinPsy thesis and will be presented to peers and staff at Lancaster University. This presentation will be available on the University's website for the public to access. There could be the opportunity to publish the findings in the Motor Neurone Disease Association newsletter and on their website, along with any other support services that have been approached. The findings will also be put forward for publication in a peer reviewed journal and/ or presented at conferences.

Ethical issues:

Confidentiality of participants will be maintained as once the survey has begun then the information provided will be anonymised meaning that no personal identifiable information will be held.

Participants will be provided with an information sheet outlining the proposed research and informed consent will be gained before the survey commences. Participants will be unable to withdraw their data once the survey has begun as all data is not identifiable at this stage, this is outlined in the consent form (see Appendix C).

The only risk of harm identified is around highlighting to participants the limitations that they are experiencing because of their diagnosis. This includes highlighting potential perceived stigma that they may be experiencing. Thus, advice and further sources of support will be provided at the end of the survey as part of the debrief procedure to ensure that participants are able to access support for any difficulties they are currently experiencing.

In terms of gaining valid consent, all participants will be aged 18 or over and will be provided with an information sheet which they will have the option of downloading and can ask questions by contacting the researcher prior to completing the survey and providing their consent. They will be unable to withdraw their data once the survey has begun as all data is not identifiable at this stage; this is outlined in the consent form (see Appendix C).

In terms of advice giving and debriefing, participants will be shown a debrief sheet following completion of the survey with the option of downloading this, which includes contact details of the researcher. They will have the opportunity to ask questions or raise concerns directly with the researcher or through the complaints procedure outlined. Further sources of support and advice will also be provided at this stage in case they require further support.

Timescale:

July – September 2017 – Submit and gain ethical approval; keep reflective diary of process; keep notes and copies of all documents relevant to process.

October – December 2017 – Conduct data collection for main study; write draft introduction and method sections; keep notes of data collection; keep reflective diary.

January – February 2018 – Analyse data; hand in draft introduction and method by end of January; write draft abstract, results and discussion.

November 2018 – December 2018 - Hand in complete first draft of research paper by end of November; keep checking for new, relevant references; complete reflective diary; produce appendices; write draft thesis abstract.

January 2019 – February 2019 – Complete final version of research paper; collate and finalise appendices; finalise thesis abstract; complete cover sheet; hand in complete draft thesis to Programme by end of March; soft-bind and hand in final thesis by deadline.

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Appendix 4-A: Study Advert

Does perceived stigma influence psychological distress in individuals with motor neurone disease/ amyotrophic lateral sclerosis/ Lou Gehrig disease?

My name is Natalie Leigh and I am conducting this research as a student in the Doctorate of Clinical Psychology programme at Lancaster University, Lancaster, United Kingdom.

What is the study about?

This study aims to find out if there is a relationship between levels of stigma and levels of psychological distress experienced by individuals with a diagnosis of motor neurone disease (MND) also known as amyotrophic lateral sclerosis (ALS) and Lou Gehrig disease. In this study, the term stigma means a feeling of shame about being different and feeling that you will be treated differently because of this, or actually being treated differently because of this difference. The study also aims to find out the strength of this relationship compared to other important factors such as symptom severity and social support.

Who can participate in this study?

Anyone aged 18 or over, who has a diagnosis of MND/ ALS/ Lou Gehrig disease and can complete an online survey regarding their experiences of stigma and well-being is eligible to take part in this study.

What will I be asked to do if I take part?

If you decide you would like to take part, you would be asked to complete an online survey to assess levels of stigma, well-being, self-esteem, social support and symptom severity. It should take you about 15 to 20 minutes to complete and need not be completed at one sitting. Someone can help you complete it if you would like them to.

Interested in taking part in this research?

If you would like to take part in this research, please click the following link to access the online survey: XXXXXXXXXXXX

Or alternatively for further information contact the main researcher: **Natalie Leigh** on n.leigh@lancaster.ac.uk or phone **07508 375657**.

Appendix 4-B: Participant Information Sheet

Participant Information Sheet

Does perceived stigma influence psychological distress in individuals with motor neurone disease?

My name is Natalie Leigh and I am conducting this research as a student in the Doctorate of Clinical Psychology programme at Lancaster University, Lancaster, United Kingdom.

What is the study about?

This study aims to find out if there is a relationship between levels of stigma and levels of psychological distress experienced by individuals with a diagnosis of motor neurone disease (MND) also known as amyotrophic lateral sclerosis (ALS) and Lou Gehrig disease. The study also aims to find out the strength of this relationship compared to other important factors such as symptom severity and social support.

Why have I been approached?

You have been approached because the study requires information from people who have MND/ ALS/ Lou Gehrig disease.

Do I have to take part?

No. It's completely up to you to decide if you take part, taking part in this research is completely voluntary. You will have the opportunity to ask questions and raise concerns at any time. If you agree to take part in the research, once the online survey has started then the information provided will be anonymised meaning that no personal identifiable information will be held. Due to this, you will not be able to withdraw your data once you have begun the survey as it will not be possible to identify your responses.

What will I be asked to do if I take part?

If you decide you would like to take part, you would be asked to complete an online survey involving questionnaires which will measure levels of stigma, levels of psychological distress, social support and symptom severity. The online survey will also request some information such as your age, gender and the time since you received your diagnosis. If you agree to take part in the research, once the online survey has started then the information provided will be anonymised meaning that no personal identifiable information will be held. Due to this, you will not be able to withdraw your data once you have begun the survey as it will not be possible to identify your responses.

Will my data be identifiable?

Once the survey has begun then the information provided will be anonymous meaning that no personal identifiable information will be held. The study findings will be produced into a report which may be published, however no personal identifiable information will be

included in the report. If a participant wants to access a copy of the results of the study then their personal details will be held long enough to send out this information, then it will be destroyed and this information will be kept confidential throughout the process. The data collected for this study will be stored securely and only the researchers conducting this study will have access to this data:

- The files on the computer will be encrypted (that is no-one other than the researchers will be able to access them) and the computer itself password protected.
- Lancaster University will keep your anonymised data for a period of 10 years after the study has finished.

What will happen to the results?

The results will be summarised and reported in a thesis and may be submitted for publication in an academic or professional journal and/ or presented at conferences. A presentation will be available on the Lancaster University website for the public to access. There could be the opportunity to publish the findings in the Motor Neurone Disease Association newsletter (in the UK) and on their website, along with any other support services that have been approached during the advertisement of this study.

Are there any risks?

There are no risks anticipated with participating in this study. However, if you experience any distress following participation you are encouraged to contact the resources provided at the end of this sheet.

Are there any benefits to taking part?

Although you may find participating interesting, there are no direct benefits in taking part.

Who has reviewed the project?

This study has been reviewed and approved by the Faculty of Health and Medicine Research Ethics Committee at Lancaster University.

Where can I obtain further information about the study if I need it?

If you have any questions about the study, please contact the main researcher:

Natalie Leigh
Doctorate in Clinical Psychology
Furness Building
Lancaster University
Lancaster, UK
LA1 4YG
Email: n.leigh@lancaster.ac.uk
Tel: +44 (0)7508 375657

Or, Research Supervisor:
Dr Fiona Eccles,
Lecturer
Division of Health Research

Furness Building
Lancaster University
Lancaster, UK
LA1 4YG
Email: f.eccles@lancaster.ac.uk
Tel: +44 (0)1524 592807

Complaints

If you wish to make a complaint or raise concerns about any aspect of this study and do not want to speak to the researcher, you can contact:

Professor William Sellwood
Tel: +44 (0)1524 593998
Email: b.sellwood@lancaster.ac.uk
Division of Health Research
Lancaster University
Lancaster, UK
LA1 4YG

If you wish to speak to someone outside of the Doctorate in Clinical Psychology Programme, you may also contact:

Professor Roger Pickup
Associate Dean for Research
Tel: +44 (0)1524 593746
Email: r.pickup@lancaster.ac.uk
Faculty of Health and Medicine (Division of Biomedical and Life Sciences)
Lancaster University
Lancaster, UK
LA1 4YG

Thank you for taking the time to read this information sheet.

Resources in the event of distress

Should you feel distressed either as a result of taking part, or in the future, the following resources may be of assistance.

1. For UK residents:

Motor Neurone Disease Association

David Niven House
10-15 Notre Dame Mews
Northampton
NN1 2BG
UK
Tel: (+44) 01604 250505
Email: enquiries@mndassociation.org
Website: www.mndassociation.org

Your local G.P. surgery can also provide advice and direct you to local services to support you in times of distress.

2. For international participants:

Australia – MND Australia

Suite 260, Level 26

100 Miller Street

North Sydney

NSW

2060

Australia

Tel: (+61) 2 8287 4980

Email: info@mndaustralia.org.au

Website: <https://www.mndaust.asn.au/Home.aspx>

Canada – ALS Canada

393 University Avenue

Suite 1701

Toronto

ON

M5G 1E6

Canada

Tel: (+1) 416 497 2267

Website: <https://www.als.ca/>

New Zealand – MND New Zealand

MND Association National Office

PO Box 24-036

Royal Oak

Auckland

1345

New Zealand

Tel: (+64) 09 624 2148

Email: admin@mnda.org.nz

Website: <https://mnda.org.nz/>

USA - ALS Association

1275 K Street NW,

Suite 250

Washington DC

20005

USA

Tel: (+1) 202-407-8580

Email: alsinfo@alsa-national.org

Website: <http://www.alsa.org/about-als/>

Appendix 4-C: Consent Form

Consent Form

Study Title: Does perceived stigma influence psychological distress in individuals with motor neurone disease?

We are asking if you would like to take part in a research project which aims to investigate whether level of perceived stigma influences psychological distress for individuals with motor neurone disease (MND, also known as amyotrophic lateral sclerosis; ALS/ Lou Gehrig disease).

Before you consent to participating in the study we ask that you read the participant information sheet and then, if you agree to continue, click each statement below to say you agree. If you have any questions or queries before signing the consent form please contact the principal investigator, Natalie Leigh on the details given on the participant information sheet.

Before proceeding to the survey, I confirm that:

- I have read the participant information sheet and understand what is expected of me within this study.
- I confirm that I understand that any responses/information I give will remain anonymous.
- I confirm that I understand that once I begin this survey that my responses will be anonymous and therefore cannot be withdrawn from the study.
- My participation is voluntary.
- I consent for the information I provide to be discussed with the researcher's supervisor at Lancaster University.
- I consent to Lancaster University keeping the anonymised data for a period of 10 years after the study has finished.
- I consent to take part in the study.

Appendix 4-D: Debrief Sheet

Debrief Sheet:

Does perceived stigma influence psychological distress in individuals with motor neurone disease?

Thank you very much your participation in this research; your time and effort has been greatly appreciated.

This research aimed to discover if there is a relationship between perceived stigma and psychological distress for people with motor neurone disease (MND) also known as amyotrophic lateral sclerosis (ALS) and Lou Gehrig disease. This also aimed to discover the strength of this relationship compared to other factors such as symptom severity and social support. It is believed that stigma will have a significant effect over and above other predictors of well-being. If results support the belief that stigma has a significant effect over and above that of other predictors of psychological distress, then this will highlight the importance of considering perceived stigma when assessing psychological distress in an individual diagnosed with MND. This will provide evidence suggesting that providing interventions aimed at reducing perceptions of stigma would be beneficial including challenging perceptions in society.

If you would like to receive the results of this study or if you have any questions then please get in contact using the details below.

Contact Details

Natalie Leigh
Doctorate in Clinical Psychology
Furness Building
Lancaster University
Lancaster
LA1 4YG
UK
Email: n.leigh@lancaster.ac.uk
Tel: +44 (0)7508 375657

Thank you for taking the time to read this debrief sheet.

Resources in the event of distress

Should you feel distressed either as a result of taking part, or in the future, the following resources may be of assistance.

1. For UK residents:

Motor Neurone Disease Association

David Niven House
10-15 Notre Dame Mews
Northampton
NN1 2BG
UK
Tel: (+44) 01604 250505
Email: enquiries@mndassociation.org
Website: www.mndassociation.org

Your local G.P. surgery can also provide advice and direct you to local services to support you in times of distress.

2. For international participants:

Australia – MND Australia

Suite 260, Level 26
100 Miller Street
North Sydney
NSW
2060
Australia
Tel: (+61) 2 8287 4980
Email: info@mndaustralia.org.au
Website: <https://www.mndaust.asn.au/Home.aspx>

Canada – ALS Canada

393 University Avenue
Suite 1701
Toronto
ON
M5G 1E6
Canada
Tel: (+1) 416 497 2267
Website: <https://www.als.ca/>

New Zealand – MND New Zealand

MND Association National Office
PO Box 24-036
Royal Oak
Auckland
1345
New Zealand
Tel: (+64) 09 624 2148
Email: admin@mnda.org.nz
Website: <https://mnda.org.nz/>

USA - ALS Association

1275 K Street NW,
Suite 250
Washington DC
20005
USA

Tel: (+1) 202-407-8580

Email: alsinfo@alsa-national.org

Website: <http://www.alsa.org/about-als/>

To download a copy of this Debrief Sheet please click on the link below:

[Debrief Sheet](#)

Appendix 4-E: Self-Report Questionnaire**Self-Report Questionnaire**

1. What is your age?

2. What is your nationality?

3. What is your gender?

4. What is your relationship status?

- a. Single
- b. Cohabiting
- c. Married/ civil partnership
- d. Divorced
- e. Widowed
- f. Other

5. How long has it been since your symptoms of motor neurone disease (MND/ ALS/ Lou Gehrig disease) began?

6. How long has it been since you were diagnosed with motor neurone disease (MND/ ALS/ Lou Gehrig disease)?

7. Have you got carer assistance in completing this survey?

Appendix 4-F: Stigma Scale for Chronic Illness (SSCI)

Please answer this on a scale of: Never, Rarely, Sometimes, Often or Always.

	Never (1)	Rarely (2)	Sometimes (3)	Often (4)	Always (5)
Because of my illness, I felt emotionally distant from other people (1)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Because of my illness, I felt left out of things (2)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Because of my illness, I felt embarrassed in social situations (3)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Because of my illness, I worried about other people's attitudes towards me (4)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I was unhappy about how my illness affected my appearance (5)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
Because of my illness, it was hard for me to stay neat and clean (6)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

Because of my illness, I worried that I was a burden to others (7)

☐☐☐☐☐

I felt embarrassed about my illness (8)

☐☐☐☐☐

I felt embarrassed because of my physical limitations (9)

☐☐☐☐☐

I felt embarrassed about my speech (10)

☐☐☐☐☐

Because of my illness, I felt different from others (11)

☐☐☐☐☐

I tended to blame myself for my problems (12)

☐☐☐☐☐

I avoided making new friends to avoid telling others about my illness (13)

☐☐☐☐☐

Because of my illness, some people seemed uncomfortable with me (14)

☐☐☐☐☐

Because of
my illness,
some people
avoided me
(15)

☐☐☐☐☐

Because of
my illness,
people were
unkind to me
(16)

☐☐☐☐☐

Because of
my illness,
people made
fun of me (17)

☐☐☐☐☐

Because of
my illness,
people
avoided
looking at me
(18)

☐☐☐☐☐

Because of
my illness,
strangers
tended to
stare at me
(19)

☐☐☐☐☐

Because of
my illness, I
was treated
unfairly by
others (20)

☐☐☐☐☐

Because of
my illness,
people tended
to ignore my
good points
(21)

☐☐☐☐☐

Some people
acted as
though it was
my fault I
have this
illness (22)

☐☐☐☐☐

People with
my illness lost
their jobs
when their
employers
found out
about it (23)

☐☐☐☐☐

I lost friends
by telling
them that I
have this
illness (24)

☐☐☐☐☐

Appendix 4-G: Depression, Anxiety and Stress Scale (DASS-21)

Please read each statement and please select an answer which indicates how much the statement applied to you over the past week. There are no right or wrong answers. Do not spend too much time on any statement.

The rating scale is as follows:

Did not apply to me at all – NEVER

Applied to me to some degree, or some of the time - SOMETIMES

Applied to me a considerable degree, or a good part of the time – OFTEN

Applied to me very much, or most of the time - ALMOST ALWAYS

	Never (1)	Sometimes (2)	Often (3)	Almost Always (4)
I found it hard to wind down (1)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I was aware of dryness of my mouth (2)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I couldn't seem to experience any positive feeling at all (3)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I experienced difficulty breathing (e.g. excessively rapid breathing, breathlessness in the absence of physical exertion) (4)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I found it difficult to work up the initiative to do things (5)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I tended to over-react to situations (6)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I experienced trembling (e.g. in the hands) (7)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
I felt that I was using a lot of nervous energy (8)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>

I was worried
about situations
in which I might
panic and make
a fool of myself
(9)

☐☐☐☐

I felt that I had
nothing to look
forward to (10)

☐☐☐☐

I found myself
getting agitated
(11)

☐☐☐☐

I found it difficult
to relax (12)

☐☐☐☐

I felt down-
hearted and blue
(13)

☐☐☐☐

I was intolerant
of anything that
kept me from
getting on with
what I was doing
(14)

☐☐☐☐

I felt I was close
to panic (15)

☐☐☐☐

I was unable to
become
enthusiastic
about anything
(16)

☐☐☐☐

I felt I wasn't
worth much as a
person (17)

☐☐☐☐

I felt that I was
rather touchy
(18)

☐☐☐☐

I was aware of
the action of my
heart in the
absence of
physical exertion
(e.g. sense of
heart rate
increase, heart
missing a beat)
(19)

☐☐☐☐

I felt scared
without any
good reason
(20)

☐☐☐☐

I felt that life was
meaningless
(21)

☐☐☐☐

Appendix 4-H: Multidimensional Scale of Perceived Social Support (MSPSS)

We are interested in how you feel about the following statements. Please read each statement carefully. Indicate how you feel about each statement using the scale below.

- 1 = Very Strongly Disagree
- 2 = Strongly Disagree
- 3 = Mildly Disagree
- 4 = Neutral
- 5 = Mildly Agree
- 6 = Strongly Agree
- 7 = Very Strongly Agree

[illegible]

My friends really try to help me. (6)							
I can count on my friends when things go wrong. (7)							
I can talk about my problems with my family. (8)							
I have friends with whom I can share my joys and sorrows. (9)							
There is a special person in my life who cares about my feelings. (10)							
My family is willing to help me make decisions. (11)							

I can talk
about my
problems
with my
friends.
(12)



Appendix 4-I: Self-Administered Amyotrophic Lateral Sclerosis Functional Rating Scale (SA-ALSFRS)

Amyotrophic lateral sclerosis (ALS) is also known as motor neurone disease (MND) and Lou Gehrig disease.

The following questions refer to how you are currently functioning at home. Please read each item carefully and base your answers on your functioning today compared to the time before you had any symptoms of ALS/ MND/ Lou Gehrig disease. Please select the response that best fits your functional status today.

Q1)

Compared to the time before you had symptoms of ALS/ MND/ Lou Gehrig disease:

Have you noticed any changes in your **speech**?

- ☐ No change (1)
 - ☐ Noticeable speech difference (2)
 - ☐ Speech has changed; asked often to repeat words or phrases (3)
 - ☐ Speech has changed; sometimes need the use of alternative communication methods (i.e. computer, writing pad, letter board or eye chart) (4)
 - ☐ Unable to communicate verbally (5)
-

Q2) Have you noticed any changes (increases) in the amount of **saliva** in your mouth (regardless of any medication use)?

- ☐ No change (1)
 - ☐ Slight but definite excess of saliva with or without night time drooling (2)
 - ☐ Moderate amounts of excessive saliva with or without minimal day time drooling (3)
 - ☐ Marked amounts of excessive saliva with some daytime drooling (4)
 - ☐ Marked excessive saliva with marked drooling requiring a constant tissue or handkerchief (5)
-

Q3) **Compared to the time before you had symptoms of ALS/ MND/ Lou Gehrig disease:**

Have there been any changes in your ability to **swallow**?

- ☐ No changes (all foods and liquids) (1)
 - ☐ Some changes in swallowing or occasional choking episodes (including coughing during swallowing) (2)
 - ☐ Unable to eat all consistencies of food and have modified the consistency of foods eaten (3)
 - ☐ Use a feeding tube (PEG) to supplement what is eaten by mouth (4)
 - ☐ Do not eat anything by mouth and receive all nutrition through a feeding tube (PEG) (5)
-

Q4) Has your **handwriting** changed? Please choose the best answer that describes your handwriting with your **dominant (usual)** hand without a cuff or brace.

- ☐ No changes (1)
 - ☐ Slower and/or sloppier but all the words are legible (2)
 - ☐ Not all words are legible (3)
 - ☐ Able to hold a pen but unable to write (4)
 - ☐ Unable to hold a pen (5)
-

Q5) The following question refers to your ability to **cut foods and handle utensils** (feed yourself) compared to before you had symptoms of ALS/ MND/ Lou Gehrig disease. If most of your nutrition is through a feeding tube (PEG), skip to **part b** of this question. If you eat most of your meals by mouth answer **part a**.

Q5a) **Compared to the time before you had symptoms of ALS/ MND/ Lou Gehrig disease:**

Cutting food and handling utensils:

- ☐ No change (1)
- ☐ Somewhat slow and clumsy (or different than before) but no assistance or adaptive equipment (2)
- ☐ Sometimes need help with cutting more difficult foods (3)
- ☐ Food must be cut by someone else but can feed slowly without assistance (4)
- ☐ Need to be fed (5)

Q5b) Using a feeding tube (PEG)

- ☐ Use PEG without assistance or difficulty (1)
 - ☐ Use PEG without assistance however may be slow and/or clumsy (2)
 - ☐ Require assistance with closures and fasteners (3)
 - ☐ Provide minimal assistance to caregiver (4)
 - ☐ Unable to perform any of the manipulations (5)
-

Q6) Compared to the time before you had symptoms of ALS/ MND/ Lou Gehrig disease:

Has your ability to **dress and perform self-care activities** (i.e. bathing, teeth brushing, shaving, combing your hair, other hygienic activities) changed?

- ☐ No change (1)
 - ☐ Perform self-care activities without assistance but with increased effort or decreased efficiency (2)
 - ☐ Require intermittent assistance or use different methods (i.e. sit down to get dressed, fasten buttons with a fastener or your non-dominant hand) (3)
 - ☐ Require daily assistance (4)
 - ☐ Do not perform self-care activities and completely dependent on caregiver (5)
-

Q7) Has your ability to **turn in bed and adjust the bed clothes** (i.e. cover yourself with the sheet or blanket) changed?

- ☐ No change (1)
 - ☐ Can turn in bed and adjust the bed clothes without assistance but it is slower or more clumsy (2)
 - ☐ Can turn in bed OR adjust the bed clothes without assistance but with great difficulty (3)
 - ☐ Can initiate turning in bed or adjusting the bed clothes but require assistance to complete the task (4)
 - ☐ Helpless in bed (5)
-

Q8) **Compared to the time before you had symptoms of ALS/ MND/ Lou Gehrig disease:**

Has your ability to **walk** changed?

- ☐ No change (1)
 - ☐ Walking has changed but do not require any assistance or devices (i.e. foot brace, cane, walker) (2)
 - ☐ Require assistance to walk (i.e. cane, walker, foot brace or hand held assistance) (3)
 - ☐ Can move legs or stand up but unable to walk from room to room (4)
 - ☐ Cannot walk or move my legs (5)
-

Q9) Has your ability to **climb stairs** changed?

- ☐ No change (1)
 - ☐ Slower (2)
 - ☐ Unsteady and/or more fatigued (3)
 - ☐ Require assistance (i.e. using the handrail, cane or person) (4)
 - ☐ Cannot climb stairs (5)
-

Q10) **Compared to the time before you had symptoms of ALS/ MND/ Lou Gehrig disease:**

Do you experience **shortness of breath** or have **difficulty breathing**?

- ☐ No change (1)
 - ☐ Shortness of breath only with walking (2)
 - ☐ Shortness of breath with minimal exertion (i.e. talking, eating, bathing or dressing) (3)
 - ☐ Shortness of breath at rest while either sitting or lying down (4)
 - ☐ Significant shortness of breath (all of the time) and considering using mechanical ventilation (5)
-

Q11) Do you experience shortness of breath or have difficulty while lying down on your back?

- ☐ No change (1)
 - ☐ Occasional shortness of breath while lying on back but don't routinely use more than two (2) pillows to sleep (2)
 - ☐ Shortness of breath while lying on back and require more than two pillows (or an equivalent) to sleep (3)
 - ☐ Can only sleep sitting up due to shortness of breath (4)
 - ☐ Require the use of respiratory (breathing) support (BiPAP® or invasive ventilation via tracheostomy) to sleep and do not sleep without it. (5)
-

Q12) **Compared to the time before you had symptoms of ALS/ MND/ Lou Gehrig disease:**

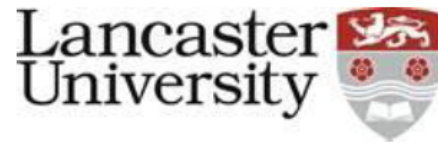
Do you require **respiratory (breathing) support**?

- ☐ No respiratory support (1)
 - ☐ Intermittent use of BiPAP® (2)
 - ☐ Continuous use of BiPAP® at night (3)
 - ☐ Continuous use of BiPAP® at night and during the day (nearly 24 hours per day) (4)
 - ☐ Mechanical ventilation by intubation or tracheostomy (5)
-

Q13) Please indicate who completed this survey:

- ☐ Patient (1)
 - ☐ Patient with assistance (2)
 - ☐ Patient with assistance from caregiver or family member (3)
 - ☐ Caregiver alone (4)
-

(BiPAP® is commonly used to describe non-invasive positive pressure ventilation and its use here in no way endorses or promotes a particular product).

Appendix 4-J: FHMREC Approval Letter

Applicant: Natalie Leigh
Supervisor: Jane Simpson and Fiona Eccles
Department: Health Research
FHMREC Reference: FHMREC17004

05 October 2017

Dear Natalie

Re: Does perceived stigma influence psychological distress in individuals with motor neurone disease?

Thank you for submitting your research ethics application for the above project for review by the **Faculty of Health and Medicine Research Ethics Committee (FHMREC)**. The application was recommended for approval by FHMREC, and on behalf of the Chair of the Committee, I can confirm that approval has been granted for this research project.

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;
- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);
- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information.

Tel:- 01542 592838

Email:- fhmresearchsupport@lancaster.ac.uk

Yours sincerely,

A handwritten signature in black ink that reads "Diane Hopkins".

Dr Diane Hopkins
Research Integrity and Governance Officer, Secretary to FHMREC.

Appendix 4-K: Ethics Amendment

An amendment was sought regarding the wording of the participant information sheet, consent form and debrief form to enhance the flow of the online survey. This also included the option for the participant information sheet and debrief sheet to be downloaded along with a change in the timescale of the project. This amendment was granted approval to be implemented on the 17th October 2017.

Appendix 4-L: FHMREC Amendment Approval Letter

Applicant: Natalie Leigh
Supervisors: Jane Simpson and Fiona Eccles
Department: Health Research
FHMREC Reference: FHMREC17027

17 October 2017

Dear Natalie

Re: Does perceived stigma influence psychological distress in individuals with motor neurone disease?

Thank you for submitting your research ethics amendment application for the above project for review by the **Faculty of Health and Medicine Research Ethics Committee (FHMREC)**. The application was recommended for approval by FHMREC, and on behalf of the Chair of the Committee, I can confirm that approval has been granted for the amendment to this research project.

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;
- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);
- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information.

Tel:- 01542 592838

Email:- fhmresearchsupport@lancaster.ac.uk

Yours sincerely,

A handwritten signature in black ink that reads "Diane Hopkins".

Dr Diane Hopkins
Research Integrity and Governance Officer, Secretary to FHMREC.